AN INVESTIGATION INTO THE EFFECTIVENESS OF PSYCHOSOCIAL SLEEP

INTERVENTIONS FOR CHILDREN WITH CYSTIC FIBROSIS

A thesis submitted in partial fulfilment of the requirements for the degree of Master of

Science in Child and Family Psychology at the University of Canterbury

By Kimberly Ballam

University of Canterbury

2023

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Acknowledgements

I would like to thank Laurie McLay and Jolene Hunter for their ongoing support and guidance throughout this process. I have been fortunate to learn from such experienced and knowledgeable researchers. Thank you both for your valuable time, patience and feedback. Thank you also to Lisa Emerson for taking the time to provide valuable feedback, to Zoe Radcliffe for assisting with the systematic review, and to Nicole Bond for the important clinical work which made this thesis possible.

To my parents, thank you for constantly motivating me. I am so grateful for the support you have both provided me during this process, including making space for me to complete my writing! Thank you also to Tracey, Jake, Michelle, Glen, and Kelly for helping me through the tough times and making sure I always had a reason to smile. I would also like to thank my friends for being so understanding and patient throughout the last year.

Thank you also to my friend Emma for her guidance while learning about cystic fibrosis. I am in admiration of your courage and the way you make the most of life. Also, to the Cystic Fibrosis New Zealand community, thank you for your assistance during recruitment and the endless work you do for New Zealanders with cystic fibrosis.

Finally, thank you to the family who participated in this research. Your efforts and commitment during this research were admirable, and this thesis would not be possible without your involvement.

Abstract

Cystic Fibrosis (CF) is an inherited and life-limiting chronic illness which affects multiple organs including the lungs and digestive system. CF can lead to many symptoms including chronic cough, poor weight gain and chest infections, which all vary in their intensity. Over the last two decades illness outcomes for those with CF have dramatically improved. Despite this, those with CF still experience significant challenges, including psychological, social and physiological difficulties, as well as a significant treatment burden.

Sleep problems are a challenge commonly reported for children with CF at a higher rate than their healthy peers. Common sleep issues include sleep related breathing disorders, reduced total sleep time, lower sleep efficiency, increased frequency of arousals, and a longer duration of wakefulness after sleep onset. This is especially problematic as sleep problems are associated with increased severity of lung disease, low mood and decreased quality of life in those with CF. Research has found that children with CF can experience sleep problems resulting from non-respiratory factors, suggesting that their sleep difficulties may be amenable to psychosocial sleep interventions.

Psychosocial sleep interventions are a category of interventions that focus on the underlying psychological or behavioural factors that impact sleep. These interventions can include cognitive behavioural techniques, sleep hygiene modifications, stimulus control techniques, sleep restriction, extinction procedures and relaxation strategies. Psychosocial sleep interventions have been found to be effective in improving sleep among typically developing children and children with neurodevelopmental disabilities, however, no research has explored the use of these interventions among children with CF. This thesis aims to investigate the use of psychosocial sleep interventions for children with CF and sleep problems through two studies.

Study one involved a systematic review of the effectiveness and acceptability of psychosocial sleep interventions among children with various Chronic Health Condition(s) (CHC), including children with respiratory conditions, diabetes and epilepsy. The review identified 13 studies which investigated the use of behavioural strategies, cognitive behavioural therapy for insomnia and motivational based interviewing techniques. The overall findings from the review were promising, with 12/13 studies reporting at least some improvement in sleep outcomes across participants. Additionally, improvements were seen in daytime behaviours, quality of life and symptoms of depression and anxiety following sleep treatment. While promising, the findings of the review are limited due to the small number of studies focused on each CHC condition, as well as methodological limitations (e.g., limited experimental control). Despite these limitations, the use of psychosocial sleep interventions emerged as a promising avenue for future research.

The findings of study one were used to inform study two. The aims of study two were to investigate the effectiveness of a psychosocial sleep intervention for a child with CF. Study two also aimed to investigate if any change in sleep would result in secondary improvements in the child and parent's wellbeing, and if the intervention would be considered acceptable to the parents. Within this study a multi-component psychosocial sleep intervention was implemented with a five-year-old boy with CF who was experiencing problematic night waking (NW) and unwanted co-sleeping. The intervention involved three sequential phases, beginning with sleep/wake rescheduling, followed by the use of reinforcement and a social story, and finally modification to parental responses to NW.

Following intervention, the frequency and duration of NW decreased significantly, which was maintained at six weeks follow up. Additionally small improvements were reported in his quality of life and daytime behaviour as measured using the Pediatric Quality of Life Inventory and Child Behavior Checklist administered pre- and post-intervention.

Some improvements were also reported in parental sleep, however, few changes were observed in regard to parental emotional wellbeing and parental relationship quality using the Depression Anxiety and Stress Scales –21 and the Relationship Quality Index. The intervention was reported be highly effective and acceptable. Despite these promising findings, further replication is needed which builds upon the limitations in existing research, including this study.

Chapter One: Introduction

Cystic fibrosis (CF) is a chronic illness which affects multiple organs, including the lungs and digestive system (Fauroux et al., 2012). Symptoms include chronic cough, shortness of breath, frequent chest infections, poor weight gain and malnutrition (Shakkottai et al., 2018). Owing to advances in research, the life expectancy and quality of life for those with CF has increased dramatically (Goetz & Ren, 2019), therefore allowing new research to focus on various areas of wellbeing. However, regardless of advances in CF research, significant challenges are still experienced by many individuals with CF, including frequent hospitalisations, mental health challenges (e.g., anxiety and depression), medical complications (e.g., CF-related diabetes), and time-consuming treatment regimens (Cystic Fibrosis NZ, n.d.).

Individuals with CF have also been found to experience high rates of sleep difficulties in comparison to their healthy peers (Fauroux et al., 2012; Naqvi et al., 2008; Vandeleur et al., 2017a; Ward et al., 2009). Specifically, those with CF tend to experience multiple sleep related breathing disorders such as nocturnal hypoxemia, nocturnal hypercapnia, and increased respiratory rates (Shakkottai et al., 2018). Those with CF may also experience poor Sleep Efficiency (SE), increased frequency of arousals during sleep, longer durations of Wakefulness After Sleep Onset (WASO), and reduced Total Sleep Time (TST; Amin et al., 2005; de Castro-Silva et al., 2010; Fauroux et al., 2012; Naqvi et al., 2008; Spicuzza et al., 2012; Suratwala et al., 2011; Vandeleur et al., 2017a & 2017b). This is concerning as adequate and restorative sleep for those with chronic illnesses is crucial for symptom management and overall health (Vandeleur et al., 2017a). Furthermore, for individuals with CF, sleep disturbance has been found to be associated with increased severity of lung disease, low mood and decreased Quality of Life (QoL; Byars et al., 2020; Fauroux et al., 2021). It is

also likely that a bi-directional relationship exists in which poor sleep exacerbates symptoms of CF, which in turn may further interfere with healthy sleep (Lewandowski et al., 2011a).

Research into sleep problems in children and adolescents (hereby referred to collectively as children) with CF has predominantly focused on medical approaches, however, emerging research shows non-respiratory factors contribute to sleep problems which may be amenable to psychosocial sleep interventions, including behavioural sleep interventions (Byars et al, 2020; Canter et al., 2021; Fauroux et al., 2021). Behavioural sleep interventions focus on making changes to aspects of the individual's sleep environment and the conditions associated with sleep onset and maintenance, in order to improve the likelihood of sleep (Sharma & Andrade, 2012). Examples of behavioural intervention strategies include sleep hygiene modifications, stimulus control techniques, sleep restriction (including the faded bedtime procedure), unmodified and modified extinction procedures and relaxation strategies (Sharma & Andrade, 2012). Although there has been little research into behavioural sleep interventions for children with CF, there is evidence to support the use of these interventions for healthy children, as well as children with other chronic health or developmental conditions (Badawy et al., 2019; Brown et al., 2013; Galland et al., 2012; Lunsford-Avery et al., 2021; Meltzer & Mindell, 2014; Park et al., 2022).

Cognitive Behavioural Therapy for Insomnia (CBT-I) is another psychosocial intervention that has been shown to be effective in treating sleep difficulties for healthy children (Paine & Gradisar, 2011; De Bruin et al., 2015; Blake et al., 2017; Ma et al., 2018; Schlarb et al., 2018), however no studies have examined its effectiveness in individuals with CF. CBT-I involves identifying and modifying unhelpful patterns of thinking and behaviour which may underly the sleep disturbance an individual experiences (Blake et al., 2017). This approach may be useful for young people with CF, as research suggests that those with CF

may experience unhelpful sleep-related cognitions and inappropriate sleep behaviours that can contribute to difficulties with sleep onset and maintenance (Canter et al., 2021).

The current study aimed to increase our understanding of the effectiveness and acceptability of psychosocial (including behavioural) sleep interventions for children with CF. To achieve this, a systematic literature review was undertaken to investigate the effectiveness and acceptability of psychosocial sleep interventions for children with Chronic Health Condition(s) (CHC) (see Chapter Two). The outcomes of this review were then used to design and evaluate a behavioural sleep intervention for a child with CF (see Chapter Three).

Cystic Fibrosis

Definition

Cystic Fibrosis (CF) is a complex and life-threatening genetic condition (Ruzal-Shapiro, 1998), which varies greatly in its severity, rate of progression and clinical expression across individuals (Castellani & Assael, 2017). CF is inherited through mutations in the gene that produce the Cystic Fibrosis Transmembrane Conductance Regulator (CFTR) protein (Cavanaugh et al., 2016). CF affects multiple organs, particularly the lungs and digestive system (Shakkottai et al., 2018). Common symptoms of CF include frequent respiratory infections, chronic cough, shortness of breath, frequent chest infections, poor weight gain and malnutrition (Shakkottai et al., 2018). One of the biggest concerns related to CF is the decline in lung functioning over time, which can subsequently result in respiratory failure owing to factors such as lower airway inflammation and recurrent respiratory infections (Shakkottai et al., 2018). Previously CF was considered fatal in infancy and childhood, however, advances in the diagnosis and treatment of CF, including improved recognition of mild symptoms, has meant that those with CF are living longer and healthier lives (Shakkottai et al., 2018). Consequently, this has allowed CF research to expand beyond

solely focusing on maintaining lung/physical health, to exploring other aspects of individuals lives, including sleep (Canter et al., 2021).

Prevalence and Trajectory

CF is also known to be more common among those of European descent, with an incidence of approximately 1 in 2000-3000 births, as opposed to an incidence of approximately 1 in 30,000 births in African, Asian and Polynesian populations (Cutting, 2015; Rafeeq & Muruad, 2017). CF is also reported to be the most common genetic illness among Caucasians (Shakkottai et al., 2018). According to the New Zealand CF Data Registry (2017), which accounts for 97-98% of all New Zealanders with CF, there are 501 registered New Zealanders with a CF diagnosis. Estimates suggest that one in every 25 Caucasian people carry the CF gene, and that approximately 1 in 625 couples in New Zealand will both carry the CFTR gene (Cystic Fibrosis NZ, n.d.). Global estimates suggest that CF affects 160,000 people across 94 countries, however approximately 57,000 of these individuals are estimated to be undiagnosed. CF is reportedly significantly undiagnosed in some countries including India, Morocco, United Arab Emirates, Oman, Japan, Argentina and Serbia (Bell et al., 2020; Guo et al., 2022). This is proposed to be due to the lack of CF research and infrastructure to support those with CF, as well as underreporting by private practitioners in these countries (Bell et al., 2020; Guo et al., 2022).

The incidence of CF has been reported to be decreasing worldwide, and the age of survival has substantially improved over the last two decades (Scotet et al., 2020). The U.S Cystic Fibrosis Patient Registry (2020) reports that the median predicted age of survival is now 59 years (95% confidence interval: 56.4-65.1 years), and within New Zealand it has been reported as 54.8 years (95% CI: 40.7 to not available; Coriati et al., 2022). This represents a significant increase from an average life expectancy of one year in 1938 when CF was first described (Scotet et al., 2020). This increase in age of survival can be attributed

to many major medical advances such as standardisation of care, improved control of pulmonary infections owing to new inhaled therapies, improved control of pseudomonas aeruginosa (bacteria that can cause infections), improved nutritional supplementation with pancreatic enzymes, lung transplantation, and implementation of genetic screening to enable earlier diagnosis (Scotet et al., 2020). In the past decade, advances in the understanding of the molecular bases of CF have also resulted in CFTR modulator therapies designed to correct the mutated CFTR protein (Scotet et al., 2020). These advances have resulted in major clinical advances in the treatment of CF, and consequently extended the lifespan of many (Cystic Fibrosis NZ, n.d.; Scotet et al., 2020).

Although the median age of survival has improved globally, various individual factors continue to impact an individual with CF's life expectancy. The most significant predictor of mortality in those with CF is poor lung function (McCarthy et al., 2015). This is assessed by measuring a person's Forced Expiratory Volume (FEV_1) from a pulmonary function test (McCarthy et al., 2015). This test calculates the amount of air an individual can force out of their lungs in one second (Hancox et al., 2021). For children, FEV_1 scores between 100% and 80% indicate mild airway obstruction, between 80% and 50% indicate moderate airway obstruction, between 50% and 30% indicate severe airway obstruction, and below 30% indicate severe airway obstruction (Jat, 2013). However, when those with CF become unwell, their FEV_1 scores are often a lot lower due to blocking of the airways (Kerem et al., 1992). This results in scores of less than 30%, which are considered to be associated with poor lung functioning and decreased age of survival (Kerem et al., 1992). Further, FEV_1 scores often decline with age, which is problematic for those with CF as these declines have consistently been associated with increases in morbidity and mortality (Szczesniak et al., 2017).

Other factors associated with a reduced age of survival include being female, the age of diagnosis (the later the diagnosis the worse the outcome), having a more severe CFTR

genotype, socio-economic status, nutritional status, pancreatic insufficiency, early colonisation of pseudomonas aeruginosa, and the presence of diabetes (Stephenson et al., 2017). Specifically for females, symptoms can become exacerbated as their lung function typically declines earlier (Ernst et al., 2010). In fact, up to the age of 20, the relative risk of survival for females compared with males has been reported to be lower (Ernst et al., 2010). This difference is proposed to be due to multiple factors, including adolescent females' difficulties with adherence to the recommended high-fat diets, as well as their reported decreases in physical activity, as opposed to adolescent males with CF (Patterson et al., 2009).

Aetiology

CF occurs as a result of a mutation in the CFTR gene, and over 2000 different types of CFTR gene mutations have been identified (Sosnay et al., 2013). The CFTR gene provides instructions for the CFTR protein, which controls how salt moves in and out of cells (Sheppard & Welsh, 1999). Variants in this gene result in CFTR protein deficiency or dysfunction that lead to abnormal fluid transportation and thickened and dehydrated mucus that impair organ function (Ley & Turck, 2022). These variants are expressed in the epithelial cells (cells which line internal and external surfaces of the body) of the lungs, pancreas, liver, intestines and sweat glands (Ley & Turck, 2022). This means mutations can result in lung infections, destruction of the pancreas, and complications in other organs (Rafeeq & Muruad, 2017). However, the specific variation to the CFTR gene will determine the CF traits an individual receives (Ley & Turck, 2022). Specifically, in the lungs, the thick mucus sticks to airway surfaces, resulting in difficulty clearing mucus, and thus increasing the risk for inflammation and infection (Ley & Turck, 2022). In the pancreas, the thickened secretions block pancreatic ducts and disrupt the transportation of digestive enzymes to the intestines,

therefore impairing the digestion of key nutrients (Ley & Turck, 2022), and creating the

potential for CF-Related Diabetes (CFRD; Ernst et al., 2010).

An individual may be born with CF when both biological parents are carriers of the

CFTR gene. It is possible that parents may not be aware they carry the gene, as having only

one copy does not affect an individual's health (Pettit & Fellner, 2014). If both parents are

carriers, but do not have CF themselves; there is a 25% chance that their child will be born

with CF. Alternatively, the child could also be a carrier but not have CF (50% likelihood), or

not be a carrier of the gene at all (25% likelihood). If an individual with CF has a child with

an individual who carries the CF gene, the child will either have a 50% likelihood of having

CF, or have a 50% likelihood of being a carrier (Pettit & Fellner, 2014).

Diagnosis and Prognosis

In New Zealand, CF is usually diagnosed soon after birth, with most babies being

screened and diagnosed before symptoms develop (Cystic Fibrosis NZ, n.d.). The Newborn

Metabolic Screening Program (also referred to as the Guthrie Heel Prick test), is often used

within the first 48 hours of life to screen babies for rare disorders including CF (Cystic

Fibrosis NZ, n.d.). Currently there is no cure for CF, however, research has begun to focus on

CFTR modifier medications, where the focus is on repairing or replacing the mutated gene,

as opposed to targeting the effects of the disease (Pettit & Fellner, 2014).

Developmental Course

Within the early years of a child with CF's life, there are significant impacts on the

parents mental wellbeing following diagnosis (Ernst et al., 2010). After a diagnosis of CF,

parents are often reported to fluctuate between normalcy and states of psychological distress,

including symptoms of depression (Glascoe et al., 2007; Ernst et al., 2010). During the first

year, some babies with CF may not show symptoms, however, frequent cough is common for

many (Goetz et al., 2019; Mayo Clinic, 2021). Importantly, during this time CF treatments

must quickly become integrated practices in the family's lives, as many adaptations are required in regard to time, finances, relationships and activities (Tluczek et al., 2015; Bell et al., 2020).

For young children with CF, language acquisition provides the opportunity to express thoughts and emotions regarding treatments, and allows children to better understand their illness (Bell et al., 2020). Additionally, the desire for independent play and increased mobility may add challenges in terms of children's willingness to engage in therapy, particularly as children become aware of the limits their illness can place on their participation in various activities (Ernst et al., 2010; Bell et al., 2020). As a child develops cognitively and emotionally, they will also likely require age-appropriate support regarding their illness, especially when they begin school and need to understand how to communicate to others what their illness means (Bell et al., 2020). Specifically, children require social support to help manage the emotional burden of their diagnosis, as well as educational support on what CF is and how to manage their symptoms and prevent illness exacerbations (Downs et al., 2006; Wong & Heriot, 2008; David et al., 2010; Jamieson et al., 2014; Bell et al., 2020).

For those with CF, adolescence can be challenging as this is when a person begins to become independent and develop their sense of identity. The desire to gain independence from parents is crucial to development, and for adolescents with CF this can mean that adolescence is the time to begin taking responsibility for treatments (Bell et al., 2020). Therefore, it is unsurprising that often during adolescence and early adulthood, consistent adherence to treatments decreases (Segal, 2008). Furthermore, physiological challenges related to puberty emerge during this time, as adolescents with more severe CF may be at risk for experiencing puberty delays (Goldsweig et al., 2019). The impact of having CF on an individual's daily life also becomes more apparent in adolescence. In a recent study, Moola et

al. (2012) investigated why youth with CF are inactive despite the importance of physical activity in preventing lung function decline. In their study of 14 youth (12 – 16 years), youth reported that their daily treatments, such as taking medications and physical therapy, reduced their time for other activities including physical activity and spending time with family and friends. Thus, adolescents with CF may face many common and illness specific challenges which impact their ability to manage their illness, and consequently impact their overall wellbeing.

During adolescence, the medical care the adolescent receives also begins to change as they transition to adult care (Nazareth & Walshaw, 2013). In New Zealand this transition begins around the age of 16 – 19 (Cystic Fibrosis NZ, n.d.). This transition results in services no longer focussing on the inclusion of parents, and instead the adolescent/young adult becomes responsible for making decisions regarding their health and treatment providers (Nazareth & Walshaw, 2013).

In adulthood, many new challenges may also emerge, including becoming parents, and finding accommodation, and financial independence (Van Gool et al., 2013; Bregnballe et al., 2017; Bell et al., 2020; Szczesniak et al., 2020). Becoming a parent may lead to increased anxiety around illness, as not only does becoming a parent often change an individual's perspective of CF (including an increase in perceived importance of minimising health decline), but also may lead to fears associated with passing the CF gene onto a child (Barker et al., 2017). Moving out from the family home can also be challenging for an individual with CF, as their new independence means they are responsible for ensuring they continue to meet their nutritional requirements, and that their living environment is healthy (i.e., avoiding second hand smoke; Bregnballe et al., 2017; Szczesniak et al., 2020).

Additionally due to the lifelong financial cost of having CF, many adults need to balance the financial demands of everyday life and the potentially large healthcare costs to

ensure access to treatments (Van Gool et al., 2013). Specifically, a study completed by Van

Gool et al. (2013) utilised the Australian Cystic Fibrosis Australia Data Registry and

estimated that the average annual healthcare cost for treating CF in Australia was USD

$15,571 (at current conversion rate equates to approximately $25,000 NZD). This cost

included CF-related medations (excluding CFTR modulator therapies), medical services,

procedures and hospitalisations (Van Gool et al., 2013). During adulthood, it is also not

uncommon that people with CF may avoid their treatments as a coping mechanism to avoid

their illness (Vélez-Vélez & Bosch, 2016), or simply because adherence to their many

recommended treatments is not possible due to the interferences treatments pose to living a

normal life (Bell et al., 2020). This can contribute to an even greater illness burden, as poor

treatment adherence has been associated with worse health outcomes and greater healthcare

use (Quittner et al., 2014).

Treatment of Cystic Fibrosis

To minimise the potential risk and impacts of CF on daytime functioning, it is

important that clinicians support those affected to maintain good mental and physical health.

This may include healthy eating, sleep and exercise (Bell et al., 2020), as well as supporting

children and young people to develop coping strategies and resilience (Quittner et al., 2008).

Additionally, individuals with CF must engage in regular time-consuming treatments each

day to manage their symptoms and maintain their physical health and functioning (Duff,

2001). Treatment routines often include frequent chest physiotherapy or exercise to promote

clearance of airway secretions and minimise the number of chest infections, as well as the use

of pancreatic enzymes with food to assist nutrient absorption, and nutritional monitoring

which often lead to the individual needing to increase their nutritional intake to between

120% and 150% of the recommended daily intake (Bell et al., 2020; Duff, 2001). Thus, the

daily lives of those with CF may revolve around managing their illness.

There are also multiple CFTR modifier medications which have emerged in recent years, including Trikafta which is a combination of three individual drugs (Elexacaftor, Tezacaftor and Ivacaftor; Zaher et al., 2021). The combination of all three of these drugs increases the activity of the CFTR protein, thus Trikafta and has been found to dramatically improve lung function and QoL, as well as lead to reductions in sweat chloride and pulmonary exacerbations (Zaher et al., 2021). Due to the extreme cost of CFTR modifier medications they are not offered in all countries. This has resulted in dramatic variation between countries for the median life expectancies of those with CF. For example, during 2018, in Poland, where no modulator therapies were available, the median life expectancy for those with CF remained at 24.5 years (Rachel et al., 2020). This is compared to a median life expectancy of 47.4 years in the USA in 2018 where modulator therapies are available, and sometimes covered by medical insurance (Link & Nayak, 2020). Fortunately, within New Zealand, Trikafta became funded by Pharmac in December 2022 (Cystic Fibrosis NZ, 2022).

Physical, Mental Health and Parental Impacts Associated with Cystic Fibrosis

Physical Health Impacts. CF affects multiple systems in the body, impacting the health and wellbeing of a person in various ways. There are many affected systems and clinical conditions, including the lungs/airways (e.g., cough, recurrent bronchitis, airway obstruction, bronchiectasis), the pancreas (e.g., steatorrhea [fatty stools], pancreatic insufficiency, CF-related diabetes), genital and urinary organs (e.g., male infertility [may be present in carriers]), and other areas (e.g., digital clubbing, early wrinkling of the skin, arthritis; Goetz & Ren, 2019, Mayo Clinic, n.d.).

The major determents reported to dictate the clinical course of CF are pulmonary exacerbations, which frequently require hospitalizations (Ernst et al., 2010). Usually for healthy individual's, when there is a build-up of mucus, coughing helps the body to clear any germs that might be trapped in mucus, however, for those with CF it is harder to cough up the

thick and sticky mucus (Fahy & Dickey, 2010). Thus, the mucus, along with any bacteria and fungus trapped in it, builds up in the lungs and means it is easier to contract viruses that can lead to inflammation, infection, chronic coughing and severe breathing difficulties (Fahy & Dickey, 2010). Consequently, the breathlessness and coughing those with CF experience can lead to an avoidance of physical activity, which in turn, further exacerbates CF symptoms (Moola et al., 2012).

This thick mucus also blocks the flow of digestive enzymes which are needed to help break down food (Ong et al., 2019). The lack of digestive enzymes can lead to nutrients not being absorbed, thus leading to poor weight gain, frequent foul-smelling bowel motions, stomach pain, excessive wind and problems with malnutrition (Cystic Fibrosis NZ, n.d.). Prior to pancreatic enzyme replacement therapy (PERT), and nutritional supplementation becoming available, nutritional failure was considered the main cause of death for infants with CF (Goetz & Ren, 2019). However despite these advances, poor nutrition is still associated with adverse clinical outcomes in CF, including decreased lung function (Steinkamp & Wiedemann, 2002). For example, a study by Konstan et al. (2003) which investigated the relationship between nutritional status and pulmonary function in children with CF, found that nutritional status at age three, predicted lung function at age six, demonstrating the interrelationship between various CF symptoms.

CF also affects the pancreas and liver, where fat malabsorption, vitamin deficiencies and protein-calorie malnutrition occur, resulting in many people with CF also developing CFRD as they age (Cystic Fibrosis Foundation Patient Registry, 2016; Elborn, 2016). CFRD results from a combination of insulin deficiency and insulin resistance, and can further impair lung functioning, and nutritional status (Moheet, 2017). Consequently, the presence of a second chronic illness (such as CFRD), adds to the already large treatment burden those with

CF face, which can further contribute to a sense of psychological distress (Collins & Reynolds, 2008).

Mental Health and Wellbeing Impacts. The wellbeing of those with CF has also been reported to fluctuate, and is impacted, at least in part, by the interaction between the severity of one's illness, treatment burdens, the care they can access, available support systems, and the individual characteristics of the person, including positive coping behaviours and resilience (Bell et al., 2020). Regardless of the severity of symptoms, those with CF, as well as their families are impacted by the awareness of the life-limiting nature of the illness (De Jong et al., 1997; Sawicki et al., 2011; Bell et al., 2020). This knowledge can result in significant negative effects on the individual's mental health and QoL (De Jong et al., 1997; Britto et al., 2002; Sawicki et al., 2011; Bell et al., 2020). Depression and anxiety have also been found to be prevalent across the lifespan for those with CF (Quittner et al., 2008; Smith et al., 2010; Modi et al., 2011; Yohannes et al., 2012; Duff et al., 2014; Fidika et al., 2014; Snell et al., 2014; Quittner et al., 2014; Cruz et al., 2009; Lord et al., 2022).

In one study by Quittner et al. (2014), the rates of depression and anxiety were investigated with a group of 1286 adolescents (*M* age 14 years) and 4749 adults (*M* age 28 years) with CF, who completed the Center for Epidemiologic Studies-Depression scale (CES-D) and the Hospital Anxiety and Depression Scale (HADS). Quittner et al. (2014) reported that elevated levels of anxiety were found in 22% of the adolescents and 32% of the adults (Quittner et al., 2014). Additionally elevated levels of depression were indicated in 10% of the adolescents and 19% of the adults who took part. Overall, levels of depression and anxiety were reported to be 2–3 times higher than those of community samples in other studies (Quittner et al., 2014), demonstrating an increased prevalence.

Vandeleur et al. (2018) also reported similar findings. Vandeleur et al. (2018) investigated the relationship between sleep quality, mood and Health Related Quality Of Life

(HRQOL) among 47 children (7 – 12 years) and 40 adolescents (13 – 18 years) with CF and sleep problems, and 55 age-matched healthy control children without CF. Age-appropriate questionnaires were used, such as to the Children's Depression Inventory (CDI) or Beck's Depression Inventory to measure mood, and the CF Questionnaire-Revised (CFQ-R) or Paediatric Quality of Life Inventory (PedsQL) to measure HRQOL. The child or adolescent completed all measures, and for the CFQ-R, both the child or adolescent and their parent completed the measure. Elevated levels of depression were indicated for 18% of the children and 20% of the adolescents with CF, as opposed to rates of only 3%, and 15% in the control children and adolescents, respectively. Additionally, no significant differences were reported between the child/adolescent and parent CFQ-R scores (Vandeleur et al., 2018). Thus, again demonstrating an increased prevalence of depression and anxiety among those with CF, as compared to their healthy peers.

Importantly, the experience of depression and anxiety can exacerbate an already challenging illness, by association with other QoL determinants such as sleep (Vandeleur et al., 2018). Depression has also been reported to be associated with worsened adherence to treatments, poorer nutritional status and decreased lung functioning among children with CF (Runions et al., 2020). While this association is clear, it is important to note that the directionality of this association is less well understood. It is likely that those with poorer physical health are at an increased risk for experiencing depression, which can create a vicious cycle which further impacts physical symptoms (National Collaborating Centre for Mental Health (UK), 2010). Furthermore, individuals with CF who experience depression are more likely to engage in risky behaviours which can worsen their symptoms, such as smoking (Quittner et al., 2008). Thus depression and anxiety can further impact the overall wellbeing of the affected individual.

Parental Impacts. Parents of children with CF are often primarily responsible for managing their child's illness exacerbations and complex daily treatments (including physiotherapy, medications and complying with dietary recommendations), which can contribute to a significant psychological strain (Grossoehme et al. 2014; Wong & Heriot, 2008; Cronly et al., 2019). These illness demands, paired with the awareness of the life-limiting nature of their child's illness, have been proposed as contributing factors toward elevated rates of depression and anxiety (Glasscoe et al., 2007; Driscoll et al., 2009; Smith et al., 2010; Besier et al., 2011; Duff, 2015; Barker & Quittner, 2016a; Cronly et al., 2019; Lord et al., 2022). For example, Quittner et al. (2014) also investigated, the rates of anxiety and depression in 3127 mothers and 975 fathers of young children with CF (M age of child = 8 years). They reported that elevated levels of anxiety were found in 48% of the mothers and 36% of the fathers (as measured on the Hospital Anxiety and Depression Scale [HADS]), and elevated levels of depression were found in 37% of the mothers and 31% of the fathers (as measured on the Center for Epidemiologic Studies-Depression scale [CES-D]).

The presence of maternal depression in mothers of children with CF has been linked to sleep problems in their children, and this relationship has also been thought to compromise the parents ability to execute necessary CF treatments, thus impacting the management of CF (Owens et al., 2002). Furthermore, multiple studies have found psychological symptoms in those with CF, as well as their parents, to be associated with decreased lung function (Ploessi et al., 2014; Cronly et al., 2019), lower body mass index (Snell et al., 2014), worse treatment adherence (Smith et al., 2010; Hilliard et al., 2015; Barker & Quittner, 2016a), worse HRQOL (Riekert et al., 2007), more frequent hospitalisations, and increased healthcare costs (Snell et al., 2014), however the direction of these associations is unclear. This is concerning as it is possible a negative cycle exists where the child's illness may contribute to impaired

psychological health for the parent, which may then contribute to a decline in the child's symptoms.

Sleep in the General Population

Sleep is an essential physiological process that is crucial to healthy development and functioning across the lifespan (Smith et al., 2021). Regardless of the presence of CF, good quality sleep can provide many benefits, and correspondingly poor sleep can have detrimental consequences. These aspects of sleep will be discussed below.

The Importance of Sleep

Sleep plays a restorative and protective role that is important for all aspects of development, including behaviour and emotional regulation, memory, learning, physical growth, and energy restoration (Richdale, 2013; Turner & Johnson, 2013). Although sleep quality is a well-recognised as a predictor for physical and mental health, there is little consensus on what constitutes "sleep quality" (Ohayon et al., 2017). Ohayon et al. (2017) conducted a review of studies in order to provide recommendations regarding indicators of good sleep quality across the life-span. Within their study it was identified that shorter sleep latencies, fewer Night Waking(s) (NW) and reduced Wakefulness After Sleep Onset (WASO) were indicators of good sleep quality regardless of age (Ohayon et al., 2017). Furthermore, higher Sleep Efficiency (SE) was reported to indicate good sleep quality across all age groups, and lower SE was indicative of poor sleep (Ohayon et al., 2017). Thus, in line with Ohayon et al. (2017)'s recommendations, it is important that indicators of good sleep quality be viewed in relation to the individual's age and needs for health and functioning.

According to Buysse (2014), a child is able to achieve adequate sleep when they have undisturbed restorative sleep of sufficient duration, that is appropriate for their age. This is defined as a multidimensional pattern of sleep-wakefulness, characterised by subjective

satisfaction, high SE (the ratio of total sleep time to time in bed; Reed et al. 2016), appropriate timing and duration, and sustained alertness during waking hours (Buysse, 2014).

There are many intrinsic and extrinsic factors that can result in sleep difficulties among children (Owens, 2020). Intrinsic factors include predisposing or contributing factors, such as the child's temperament, medical issues, neurodevelopmental disabilities, or anxiety disorders, whereas extrinsic factors may include characteristics of the parents or the environment which interfere with the child's ability to sleep (Owens, 2020).

Sleep Problems in Typically Developing Children

Unfortunately many children do not obtain sufficient sleep quantity and quality for their age (Owens et al., 2014; Becker et al., 2015). In fact, research indicates that more than a quarter of children will experience sleep difficulties at some point in their childhood (Owens, 2008). These rates are even higher among children with complex support needs, such as children with psychiatric, developmental and/or medical conditions (Meltzer & Mindell, 2008).

The most common type of childhood sleep problem is insomnias, which involve problematic sleep quality, timing and duration, which are often reflected in difficulties initiating and maintaining sleep (Roth & Roehrs, 2003; Krystal, 2005; American Psychiatric Association, 2013; Ohayon et al., 2017). Such sleep problems can present in a variety of ways for children, including bedtime resistance, sleep onset delay (i.e., the inability to fall asleep within an age-appropriate amount of time), Early Morning Waking(s) (EMW), and frequent and prolonged NW (Owens et al., 2022). These difficulties can result in reduced Total Sleep Time (TST), reduced sleep quality and fragmented sleep (Owens et al., 2022).

For young children and their parents, sleep onset delay and frequent NW are two of the most common sleep difficulties, and have been reported to occur in 20–30% of children under the age of three (Sadeh et al., 2009). NW are characterised by caregiver complaints,

however are often a normal part of sleep architecture at this age (Moore, 2007). For children under three who experience difficulties with sleep onset and NW, they are often unable to return to sleep independently following NW (Moore, 2007). The reasons for these difficulties will be discussed in a later section of this chapter.

Acts of bedtime resistance (e.g. bedtime avoidance, refusal during the bedtime routine, frequent problematic requests following being put to bed), while developmentally normal, are often the parents first experiences of having their parental limits tested (Moore, 2007). These behaviours often emerge following inconsistent sleep rules and routines, or when the child dictates routines (e.g. the child decides when they want to go to bed and/or determines all aspects of the bedtime routine; Moore, 2007). This can lead to conflict between the child and parent due to differing desires, and in some cases, exacerbate sleep onset delays when not dealt with appropriately (Freeman, 2006; Moore, 2007; Flores et al., 2016). Furthermore, bedtime resistance has been associated with various sleep problems including NW, sleepwalking and difficulty waking children in the morning (Blader et al., 1997; Wilson et al., 2014).

Children also frequently experience nightmares, with research suggesting that 75% of kindergarten children have experienced at least one bad dream or nightmare (Mindell & Barrett, 2002). At age nine, nightmares and bad dreams are reported to peak, however during the ages of 10 – 12, they then tend to decrease in frequency (Muris et al., 2001; Moore, 2007). The content of bad dreams has been reported to often relate to the child's specific developmental stage, with imaginary creatures being the most common content among children aged 7 – 9, and being kidnapped being most common among children aged 10 – 12 (Moore, 2007; Muris et al., 2000). Nightmares, night-time fears and bad dreams each have distinguishing features. Nightmares can be distinguished from bad dreams as they typically lead to the child waking (American Academy of Sleep Medicine, 2005), and night-time fears

are experienced while the child is awake (Moore, 2007). Night-time fears have also been found to be more common than nightmares (Muris et al., 2000), and it has been reported that night-time fears are experienced by nearly 80% of school aged children (Gordon et al., 2007; Muris et al., 2001).

Parasomnias (e.g., sleep terrors, sleepwalking, nightmare disorder, sleep-related eating disorder and sleep paralysis) are disruptive sleep-related disorders that involve incomplete waking from Non Rapid Eye Movement (NREM) sleep (Howell, 2012). These can occur during different stages of sleep and are often accompanied by alterations in emotions and perceptions (Biolodeau et al., 2019). Parasomnias are reported to be relatively common during childhood, with estimates of up to 78% of children aged three to 13 experiencing one parasomnia, however these often disappear in adolescence (Laberge et al., 2013; Biolodeau et al., 2019). Children may be at risk for experiencing parasomnias due to exposure to stressful events and/or individual characteristics, including the presence of anxiety, depression, hyperactivity-inattention, and other sleep difficulties (Sadeh, 1996; Stein et al., 2001; Laberge et al., 2013; Biolodeau et al., 2019). Nightmares and bad dreams can often easily be distinguished from parasomnias (i.e., sleep terrors), as during a bad dream or nightmare a child can remember the content of the dream/nightmare, whereas for a sleep terror children do not remember what has occurred (American Academy of Sleep Medicine, 2005; Moore, 2007).

EMW are another issue commonly experienced by children (Jenni & LeBourgeois, 2006). Although what is considered "waking too early" is dependent on the specific family's needs, problems with EMW have been previously defined as the child frequently waking before 5.00am (Wiggs & Stores, 1998). Additionally, determining the presence of EMW can further be complicated when it is unclear if the EMW is a NW or represents the end of the child's sleep (Meltzer, 2010). It is also important to differentiate between the child simply

having a morning circadian preference, or if EMW are occurring due to bedtimes that are too early and/or sleep interfering behaviours, such as the presence of napping during the day (Meltzer, 2010).

Co-sleeping may also occur during childhood which is defined as a parent sleeping in the same space as their child (Dodd & Jackiewicz, 2015). Although co-sleeping is not necessarily considered a problem due to cultural preferences, societal values and/or personal preferences (Keller & Goldberg, 2004; Goldberg & Keller, 2007), it can become problematic when it is unwanted by the parents. Unwanted co-sleeping can be parent- or child-initiated and occurs when despite parents preference to sleep separately, the parent and child sleep in the same space (i.e., in the child or parents' bedroom; McLay et al., 2018). This often occurs as an attempt to resolve the child's difficulties with initiating and maintaining sleep (Keller & Goldberg, 2004; Goldberg & Keller, 2007; Ramos et al., 2007; McLay et al., 2018). Although conflicting perspectives on the advantages and disadvantages of co-sleeping exist, many believe that co-sleeping can lead to sleep problems in children as it promotes problematic sleep-onset associations that do not encourage independence (Ramos et al., 2007; Cortesi et al., 2008).

Another common sleep disorder, particularly during adolescence, is delayed sleep phase syndrome, which is estimated to impact 5-10% of adolescents (Crowley et al., 2007; Moore, 2007). Delayed sleep phase syndrome involves significantly delayed sleep onset times (of at least two hours) compared to the individual's desired bedtime, as well as extended Sleep Onset Latency (SOL) when attempting to fall asleep at more conventional bedtimes (Moore, 2007; Micic et al., 2016). This can occur during adolescence as a result of a biological delay in the circadian rhythm which leads to a slower build-up of sleep pressure, thus delaying the time an adolescent becomes tired in the evening (Sivertson et al., 2013). Furthermore, late sleep onsets, paired with the physiological need for adequate sleep can

result in difficulty waking in the morning, which can thus lead to later waking's that reinforce the delay in building of sleep pressure (Sivertson et al., 2013; Micic et al., 2016). When this pattern of insufficient sleep occurs over multiple nights, the presence of an excessive sleep need builds, which can result in disruptions to daily functioning (e.g. cognitive functioning, daytime sleepiness, irritable mood, and impacts on school and extracurricular activity attendance, Crowley et al., 2007; Moore, 2007; Van Maanen et al., 2013; Micic et al., 2016).

Cystic Fibrosis and Sleep Problems

Sleep problems are a common co-occurring problem among children with CF, with estimates suggesting that subjective sleep problems exist in more than 50% of those with CF (Fauroux et al., 2012; Naqvi et al., 2008; Ward et al., 2009). Research into sleep problems in children with CF is understudied in comparison to adult populations with CF (Canter et al., 2021), and most studies on sleep problems in children are in older youth with lung dysfunction (Byars et al., 2020). As such, the true prevalence of sleep disturbance across younger age groups with CF is not known (Byars et al., 2020). It is also important to note that sleep difficulties for those with CF may be reported less frequently than for typically developing individual's (Lewandowski et al., 2011a). This may be due to those with CF viewing their sleep problems as less significant than their illness-related symptoms, or because they assume that the sleep problem is an inevitable and untreatable part of their illness (Quine, 1992). Despite the lack of knowledge on sleep problems among children with CF, it is recognised that CF-related symptoms and common sleep challenges contribute to children with CF experiencing an increased risk of both physiological and/or behavioural sleep difficulties (Canter et al., 2022b).

Research into CF and sleep demonstrates that children with CF tend to report poor subjective sleep (Amin et al., 2005; Naqvi et al., 2008; Fauroux et al., 2012; Flume et al., 2009; Cavanaugh et al., 2016; Vandeleur et al., 2017a; Byars et al., 2020; Louis et al., 2022).

This has been attributed to multiple factors including the presence of nocturnal coughing, pain, gastroesophageal reflux, and medication side effects (Fauroux et al., 2012; Flume et al., 2009; Louis et al., 2022). Furthermore, studies have found that sleep problems in children with CF are common and often related to illness factors. For example, a study by Vandeleur et al. (2017a) investigated sleep quality and sleep patterns among clinically stable children with CF and healthy control children (aged 7 – 18 years). They reported that even in periods of clinical stability, the children with CF were found to have less TST than their peers. This difference was reported by Vandeleur et al. (2017a) to be accounted for by children with CF experiencing more time awake during the night, rather than less time spent in bed. Furthermore, despite the children being considered clinically stable, objective sleep measures (actigraphy) demonstrated that sleep quality, SE, TST, the frequency of NW, and WASO, were related to illness severity as measured by FEV_1.

A study by Naqvi et al. (2008) also demonstrated the relationship between sleep difficulties and lung functioning in children with CF, paying attention to sleep difficulties that were not addressed in the Vandeleur et al. (2017a) study. Naqvi et al. (2008) looked at the sleep architecture of 24 children with CF and 14 healthy controls (*M* age 14 years) by using questionnaires and an overnight polysomnography assessment. They found that 43.5% of those with CF reported sleep onset problems, 30.4% snored at night, 39.1% had difficulty with sleep maintenance, and 73.9% reported daytime sleepiness. Additionally, those with CF experienced a significant decrease in SE, prolonged Rapid Eye Movement (REM) latency, and a reduction in the percentage of total REM sleep. There was also an association discovered between SE and FEV_1 suggesting that sleep quality in children and adolescents with CF is expected to be disrupted by their disease or vice versa.

Another study by Amin et al. (2005), also demonstrated the relationship between lung functioning and impaired sleep in a larger population of children. Amin et al. (2005)

investigated the relationship between sleep disturbance and pulmonary function in 40 children with stable CF and 40 age -matched controls (aged 8 – 18 years). They found that those with CF had significantly lower SE than control subjects, and the FEV1 of those with CF correlated positively with sleep duration and SE. Additionally, FEV1 scores correlated negatively with the number and duration of NW, and the age and Body Mass Index (BMI) of the child. Amin et al. (2005) also reported that those with CF had more NW and an average TST of 24 minutes less than healthy controls, therefore further exemplifying the relationship between problematic sleep and worse illness outcomes in children with CF.

This relationship between illness severity and sleep has also been demonstrated qualitatively in children and adults with CF. Specially a study by Stenekes et al. (2009) investigated the frequency, severity and self-management of pain, shortness of breath and cough in 123 children and adults with CF through a mailed survey. Their results indicated that 63% of the children and adults with CF associated coughing with sleep disruption. Additionally, Stenekes et al. (2009) found an association between worse reported FEV1 scores and sleep duration and efficiency.

Importantly various other studies have also reported that children with CF have sleep durations that are significantly less than their healthy peers (Silva et al., 2016; Waters et al., 2017; Fauroux et al., 2012; Vandeleur et al., 2017a; de Castro-Silva et al., 2010; Suratwala et al., 2011; Meltzer et al., 2012; Paranjape et al., 2015; Ramos et al., 2011). This is concerning as obtaining enough sleep is essential for healthy development and functioning regardless of the presence of CF (Smith et al., 2022).

A study by Meltzer et al. (2012) demonstrated this as they investigated the sleep patterns and associations between sleep and perceived health for 45 children with CF and 45 healthy developing children (aged 3-5 years) through parent-report questionnaires. Their findings demonstrated significant differences in the sleep patterns, and presence of symptoms

of SDB and sleep difficulties between the two groups, with the children with CF having worse sleep (Meltzer et al., 2012). They also discovered an association between poorer perceived health and sleep disturbances among the children with CF, but not for the healthy children. Furthermore, they reported that the children with CF slept an average of 42 minutes less than their healthy peers, which was explained by later bedtimes (by 30 minutes). Thus, many children with CF experience sleep difficulties which are associated with worse perceptions of their illness or worse physiological outcomes.

Older children with CF are also at risk of experiencing insomnia which may put them at risk for mental health difficulties. For example, in a study by Tomaszek et al. (2018) the impacts of insomnia on the QoL of adolescents and adults with CF were investigated. Tomaszeck et al. (2018) provided a questionnaire to 95 adolescents and adults with CF (aged 14 – 25 years) and found that insomnia was diagnosed in 38% of the respondents. Furthermore, the risk of insomnia increased with increases in anxiety and depressive symptoms, and insomnia was found to significantly worsen QoL reports (Tomaszek et al., 2018).

Contributing Factors to Poor Sleep in Children with Cystic Fibrosis

There are many potential factors which influence the quality and quantity of sleep a person obtains. For those with CF in particular, there are a multitude of physiological and behavioural factors that may play a role. Evidence suggests there may be an interaction between sleep problems and CF-related symptoms, where sleep problems may exacerbate CF-related symptoms, and this exacerbation in symptoms can in turn, disrupt sleep (Vandeleur et al., 2017b). Furthermore, the physical, social and emotional demands of a chronic illness often contribute to further barriers to obtaining quality sleep, therefore these factors should be considered in assessment and treatment planning (Canter et al., 2021).

Physiological Factors. During sleep there are many physiological changes to breathing patterns which occur between infancy and adulthood. Specifically, after birth, more stable breathing patterns begin to emerge, which are then followed by a decrease in respiratory rate (number of breath's per minute) as individuals age (Al-Hathlol et al., 2000). With increasing age, there is also a decrease in ventilation, and an increase in tidal volume (amount of air a person inhales during a breath; Black-Pearlman, 2007; Al-Hathlol et al., 2000). The pattern of respiration also changes throughout life, with inspiratory time (time taken for inhalation; Parthasarathy et al., 2000) increasing threefold from preterm to adulthood, and expiratory time correspondingly decreasing twofold (MacLean et al., 2015).

For those with CF, sleep architecture is disrupted as the normal changes in ventilation which occur during sleep are magnified (Marcus, 2001). Those with a low Functional Residual Capacity (FRC; i.e. the volume remaining in the lungs after a normal exhalation; Hopkins & Sharma, 2021), have little functional reserve, thus their lungs are more likely to desaturate (Hopkins, 2021). This desaturation occurs due to both REM-related intercostal muscle hypotonia (i.e., poor muscle tone in the muscles that form and move the chest wall; Madhok & Shabbir, 2022), and an increased ventilation–perfusion mismatch (i.e., mismatch between blood flow and ventilation in the lungs; Tsang & Hogg, 2014; Marcus, 2001). Studies have demonstrated sleep-related desaturation in paediatric patients with CF (Coffey et al., 1991; Spier et al., 1984).

During sleep, those with CF may also experience coughing, gastrointestinal discomfort, sinus congestion and wheezing, leading to NW (Canter et al., 2021). This is because the regulation of respiration during sleep differs significantly from wakefulness (Malik et al., 2012). These changes occur as there is no longer input from the waking state to regulate breathing (Malik et al., 2012). It is therefore unsurprising that all respiratory disorders are worse during periods of sleep when compared to wakefulness (Marcus, 2001).

Although there is limited research on sleep in children with CF, there are multiple studies focusing on adults. These studies have demonstrated several co-occurring features associated with sleep disturbance, including chronic pain (Flume et al., 2009), low mood (Dancey et al., 2002), iron deficiency (Hayes, 2007), asthma, Gastroesophageal Reflux Disease (GERD) and Percutaneous Endoscopic Gastrostomy (PEG) feeds (Katz, 2014). These co-occurring conditions can affect sleep either alone or in combination, thus, there is not one singular clinical feature which solely contributes to sleep disturbance in those with CF.

Other physical conditions that may disrupt sleep for those with CF include nocturnal hypoxemia, nocturnal hypercapnia, increased respiratory rate and obstructive sleep apnea, which are discussed below.

Nocturnal Hypoxemia. Individuals with CF often experience nocturnal hypoxemia, which is a sign of breathing or circulation problems (Bhutta et al., 2022). Nocturnal hypoxemia occurs when there is a below-normal level of oxygen in an individual's blood (Milross et al., 2001). Children with moderate to severe CF lung disease have been observed to experience clinically significant nocturnal hypoxemia, which is thought to occur as a result of sleep-related reductions in lung volumes and ventilation (Tepper et al., 1983; de Castro-Silva et al., 2009; Ramos et al., 2013). Children with CF, including those who have no clinical evidence of lung disease, have also been reported to experience a higher frequency of desaturation episodes and lower nocturnal oxygen saturations as compared to healthy children, despite having normal daytime saturations (Naqvi et al., 2008; Vandeleur et al., 2017b; 2017; Suratwala et al., 2011; Spicuzza et al., 2012; de Castro-Silva et al., 2009; van der Giessen et al., 2012; Shakkottai et al., 2018). Nocturnal hypoxemia has also been found to be associated with sleep difficulties among children and adults with CF, including poor

subjective sleep quality, and reduced SE and TST (de Castro-Silva et al., 2009; Perin et al., 2012; Milross et al., 2002; Vandeleur et al., 2017b; Dancey et al., 2002).

Nocturnal Hypercapnia. Individuals with CF may also experience nocturnal hypercapnia, which occurs when there is too much carbon dioxide (CO_2) in the bloodstream due to impaired ventilation during sleep (Waters et al., 2017; Fauroux et al., 2012; Suratwala et al., 2011; Perin et al., 2012; Tepper et al., 1983; Milross et al., 2001). Hypercapnia occurs when the body does not receive enough fresh oxygen, or does not get rid of enough CO_2; thus, a person may need to gasp to balance their levels of oxygen and CO_2 (Patel et al., 2002). Nocturnal hypercapnia has been found to be associated with poor subjective sleep quality (Fauroux et al., 2012; Milross et al., 2002), low lung function (Waters et al., 2017; Perin et al., 2012; Milross et al., 2001), and reduced nocturnal oxygen saturations (Silva et al., 2016; Perin et al., 2012) in those with CF.

Increased Respiratory Rate. Compared to healthy controls, individuals with CF may also experience an increased respiratory rate (e.g., shallow rapid breathing) during sleep (Paranjape et al., 2015; Bradley et al., 1999; Young et al., 2008). This is important as Perin et al. (2012) documented that adults with CF who while awake experience fast breathing, tend to experience lower nocturnal oxygen saturations and worse hypercapnia. Thus, if this pattern continues during sleep, those with CF may be at greater risk of experiencing nocturnal hypoxemia or hypercapnia, further worsening the quality of their sleep. Waters et al. (2017) also found that there was an association between increased respiratory rate during sleep and a lower FEV1 score and worsened nutritional status for children with CF.

Sleep Disordered Breathing. Those with CF may also experience problems of Sleep Disordered Breathing (SDB), such as Obstructive Sleep Apnea (OSA). OSA is characterised by repeated cessations of airflow during sleep which last up to ten seconds (Xie, 2011). This occurs due to the obstruction of the upper airway, regardless of adequate continued

respiratory effort (Xie, 2011). OSA affects many people regardless of the presence of CF, however there appears to be a correlation between the severity of CF lung disease, and nocturnal oxygen saturations (Reiter et al., 2021). The frequency of OSA has also been reported to be higher among young children with CF compared to healthy control children (Spicuzza et al., 2012; Ramos et al., 2009; Ramos et al., 2011; Veronezi et al., 2015). According to polysomnographic data, it is also estimated that 55%–75% of children with CF experience OSA (Shakkottai et al., 2022). Furthermore, in comparison to healthy controls, children and adults with CF who have been referred to sleep-laboratories are three times more likely to experience moderate to severe OSA (Shakkottai et al., 2022).

Children with CF who have OSA have also been found to have lower nocturnal oxygen saturations, as compared to children with CF who do not have OSA (Waters et al., 2017; Spicuzza et al., 2012; Ramos et al., 2013; Ramos et al., 2009; Ramos et al., 2011), illustrating the cumulative impact that multiple physical symptoms may have on sleep quality. The most common SDB complaint for children with CF is snoring, which is followed by difficulty breathing during the night and daytime sleepiness (Reiter et al., 2021; Shakkottai et al., 2022). Interestingly, SE and the frequency of arousals may not differ in children with CF who do and do not have OSA (Ramos et al., 2009). This raises the possibility that regardless of the high rates of SDB among children with CF, OSA may not be the sole cause of sleep complaints (Shakkottai et al., 2018).

Circadian Rhythm Disturbances. All individuals have circadian rhythms which follow a 24-hour pattern, and for humans these are regulated by the Suprachiasmatic Nucleus (SCN) of the anterior hypothalamus (Kondratova & Kondratov, 2012). Zeitgebers, which are environmental cues (e.g., light, social interactions and mealtimes), regulate the SCN and consequently impact underlying circadian timings (Baron & Reid, 2014). Circadian rhythms are critical for healthy sleep as research has shown that desynchrony between internal

circadian timing systems and sleep–wake times may disrupt various physiological systems (Reddy et al., 2022). In regard to CF, it has recently been discovered that CFTR dysfunction may lead to circadian rhythm disturbances and consequently impact sleep (Louis et al., 2022). For example, research has suggested that the CFTR gene may contribute to regulating circadian rhythms (Louis et al., 2022). In one study, Barbato et al. (2019) compared tissue samples from CF-afflicted and healthy mice in both standard and sleep-deprived states, and found a significant expression of dysregulated circadian clock genes among CF mice, which the authors attributed to a loss of CFTR functionality. Thus, it is possible that there is a CF related genetic contribution to the sleep problems those with CF experience.

   Chronic Pain. Sleep quality for those with CF may also be impaired due to chronic pain (Reiter et al., 2021). Specifically, two-thirds of children with CF experience recurrent pain episodes, including abdominal, musculoskeletal and joint pain (Reiter et al., 2021). Research has found that poor sleep can lead to higher perceptions of pain symptoms (Allen et al., 2016), thus a bidirectional relationship may exist where pain leads to poor sleep, and poor sleep leads to experiencing higher pain levels (Roehrs & Roth, 2005). Although many studies have linked pain and sleep problems in those with CF (Allen et al., 2016), previous research has not examined the impact of improving sleep quality on pain in children with CF (Reiter et al., 2021).

   Environmental and Behavioural Factors. It is important to understand that children of all ages with CF may struggle with time-consuming treatments, sleep disrupting symptoms, and anxiety around their illness, which all can have substantial negative impacts on sleep (Vandeleur et al., 2017a). In their study examining the relationship between sleep quality and CF severity among clinically stable children with CF and healthy control children, Vandeleur et al. (2017a) reported that one of the most observable explanations for the difference in TST between those with CF and their healthy peers was the time needed for

various treatments, such as physiotherapy and nebulised medications. These findings are supported by a qualitative study from Canter et al. (2021), in which youth (aged 11 – 17 years) with CF were asked about their sleep habits, CF-related concerns and perspectives on potential behavioural sleep intervention strategies. In their study, several youth reported having to wake up early or stay up late in order to complete necessary airway clearance routines, thus demonstrating environmental factors which challenge a child's ability to obtain adequate sleep.

Although the time taken to complete CF-related treatments is significant across ages, there may be a difference in the impacts on TST during different stages of childhood (Vandeleur et al., 2017a). For example, Vandeleur et al. (2017a) suggested that the time taken to perform CF treatments may interfere with sleep time in preschool children, however by school age, later bedtimes allow more time for treatments without leading to sleep loss. Nonetheless, regardless of age it is important to note that as reported earlier, children with CF experience significantly less TST than what is recommended (Paruthi et al., 2016).

Further, another study by Vandeleur et al. (2017b) highlighted the importance of rectifying these sleep difficulties in children with CF, as they investigated the clinical correlates of sleep disturbance in children with CF. Vandeleur et al. (2017b) found that those with impaired lung function ($FEV_1 = 80\%$), frequent nocturnal cough, behaviour disorders, a diagnosis of asthma, lowered baseline oxygen saturation during sleep, those requiring nocturnal PEG feeds, and those with non-respiratory comorbidities such as CFRD, had significantly more sleep disturbance than the children without these conditions.

Those with CF may also experience anxiety around their illness, which can also impact their sleep. Further, there is evidence of high rates of anxiety in those with CF (Bell et al., 2020), and a known link between anxiety and sleep problems (APA, 2013). Therefore, a negative cycle may exist where anxiety could be interfering with sleep, however also be

worsened as a result of sleep deprivation. Thus, the link between CF, sleep and mental health is likely to be bidirectional.

Another factor that may impact sleep in those with CF is irregularities in their sleep environment, owing to frequent hospital admissions (Canter et al., 2021). Research suggests that an unfamiliar and disruptive hospital environment can result in frequent NW, as well as family separation and disruptions to daily routines, all which can impair an individual's sleep (Boman et al., 2003; Dogan et al., 2005; Hinds et al., 2007; Jacob et al., 2006; Jarman et al., 2002; Walker et al., 2010; Whitsett et al., 2008; Zhou et al., 2017). For example, in a study by Jacob et al. (2007), 25% of hospitalised children with cancer reported sleep fragmentation and NW. Furthermore, sleep disturbances for those with CF may also occur due to specific treatment regimens, disruptions in routine, frequent interruptions by medical professionals, fears and anxiety around the illness, separation from parents, or a combination of such factors (Lewandowski et al., 2011a).

Consequences of Poor Sleep for Children with Cystic Fibrosis

Regardless of the presence of a chronic illness, inadequate sleep quality and quantity are associated with a wide range of negative consequences for children, including daytime sleepiness, inattention, cognitive deficits (Beebe, 2011), school attendance problems (Gibson et al., 2006, Dewald, Meijer, Oort, Kerkhof, & Bögels, 2010), and an increased risk of emotional and behaviour difficulties (Chorney et al., 2008; Curcio et al., 2006; Paavonen et al., 2016).

Physical Consequences of Sleep Problems. Poor sleep is known to have multiple detrimental effects on physical health, such as increasing susceptibility to infections (e.g., the common cold; Irwin, 2015; Shakkottai et al., 2018) and impairing immune system functioning and metabolic/endocrine regulation (Bryant et al., 2004). Sleep disturbances are also associated with various adverse cardiovascular, immune, and metabolic outcomes which

may impact illness severity for those with CF (Bryant et al., 2004; Waters et al., 2013). Specifically, individual's with CF are at risk for CFRD and recurrent respiratory tract infections, which may be exacerbated by poor sleep (Vandeleur et al., 2017a). However, to date, very little research has investigated the physical consequences of poor sleep for those with CF (Reiter et al., 2021).

The association between poor sleep and obesity is also significant for those with CF, as maintaining adequate physical health and diet is paramount to CF management (Harindhanavudhi et al., 2020). Importantly, obesity is considered one of the most significant risk factors for sleep disturbances in healthy individual's, as obesity can increase the risk of developing OSA (Muscogiuri et al., 2019). Likewise, poor sleep quality may also contribute to obesity, and several studies have identified sleep difficulties as a predisposing risk factor for obesity (Hargens et al., 2013; Lyytikainen et al., 2011; Markwald et al., 2013). Specifically, short sleep durations (between 5 – 6 hours) have been found to increase the risk of developing obesity (Alodhayani et al., 2017; Broussard & Van Cauter, 2016; Canuto et al., 2014), and the association between the quality and quantity of sleep with body mass is well established (Buxton & Marcelli, 2010; Grandner et al., 2012; Rao et al., 2009).

One study by Hanna and Weiner (2014) investigated the variations in weight in 226 children with CF (aged 2 – 18 years) using data from the U.S CF Patient Registry. In their study nutritional status was classified in accordance with the CF Foundation guidelines, and of the 226 patients with CF, 57% had a BMI which was considered within a healthy weight status, 7% were in nutritional failure, 12% were at risk of nutritional failure, 15% were overweight, and 8% were obese. Although these rates of obesity and being overweight are lower than what are reported in the overall population, (28.7% of adults in New Zealand were considered obese in 2013/2014; Ministry of Health, 2014), the low rates could reflect additional effort by those with CF to ensure they maintain adequate nutrition. Thus it is

important to consider how lifestyle factors such as maintaining a healthy BMI, may further impact an individual with CF's sleep.

Mental Health and Wellbeing Consequences of Sleep Problems. In addition to the many physical health implications associated with sleep difficulties, sleep problems have also been associated with mental health and wellbeing difficulties among those with CF, however the direction of this association has not been confirmed. For children and adults with CF, impaired sleep quality has been found to be associated with poorer HRQOL (as measured by the CFQ-R, World Health Organization Quality of Life [HOQOL-BREF], and Cystic Fibrosis Quality of Life [CFQOL]) and social and emotional functioning (Dancey et al., 2002; Bouka et al., 2012; Forte et al., 2015; Íscar-Urrutia et al., 2018; Shakkottai et al., 2018; Tomaszeck et al., 2018; Vandeleur et al., 2018).

In the study by Vandeleur et al. (2018) discussed earlier, the authors also investigated the associations between sleep quality, HRQOL and mood. They reported that not only did the children with CF report poorer sleep quality than the controls, but their sleep quality (as measured by overnight oximetry, actigraphy) and daytime sleepiness (as measured by the Pediatric Daytime Sleepiness Scale) were also associated with self-reported lower mood (as measured by the CDI or Beck's Depression Inventory) and reduced HRQOL (as measured by CFQ-R or PedsQL). This association differed with age, however poor sleep quality and greater sleepiness were observed across all ages. In younger children (7 – 12 years) there was a negative correlation between mood score and SE, as well as sleepiness scores being predictive of mood score. However, in the older children (13 – 18 years) poorer sleep and more sleepiness were predictive of lower HRQOL, but not mood. Furthermore, there were no associations between sleep, mood or HRQOL in the control children. Thus, in children with CF, despite sleep difficulties being associated with worse HRQOL, it is likely there is an age-

specific manner to the impacts on mood, which may be explained by various extraneous factors.

Interestingly, different findings were reported by Cavanaugh et al. (2016) who investigated the associations between poor sleep, behaviour and QoL in 50 children with CF (aged 6-19 years) using actigraphy, the Behavior Assessment System (BAS), and the CFQ-R. Their findings indicated that 80% of children experienced poor sleep, and that lower SE was associated with more attention and hyperactivity problems. Additionally, WASO was associated with worsened attitudes towards teachers and attention problems, however there were no associations found between sleep and QoL or pulmonary functioning (Cavanaugh et al., 2016). Therefore, their findings suggest that different sleep outcomes (e.g., SE and WASO) may be associated with attention and hyperactivity problems and worsened attitudes towards teachers, but not QoL or lung functioning. Interestingly, the children in this study were however reported to have relatively normal pulmonary functioning (as measured by FEV1), which may have contributed to the lack of statistically significant relationship found between sleep characteristics and pulmonary functioning. Although both the study by Vandeleur et al. (2018) and by Cavanaugh et al. (2016) were similar in their methodologies, statistical analysis and measures used, the study by Cavanaugh et al. (2016) was limited by not including a comparison group of healthy control children, however this does not explain the differences in findings between the two studies. Overall, these differing findings therefore illustrate that there is little consensus on the impacts poor sleep can have on mental health and wellbeing among children with CF, thus it is important that further research be completed.

Relationship Between Poor Sleep and Family Characteristics

There is a well-established relationship between sleep problems in children with CF and family characteristics, including parental stress, depression, anxiety and sleep (Sivertsen

et al., 2009; McQuillan et al., 2019; Byars et al., 2020). As previously mentioned, research suggests that parents of children with CF may be at risk of developing mental health difficulties and experiencing elevated levels of family stress (Hiscock & Wake, 2002; Meltzer & Mindell, 2007; Mindell et al., 2011; Thome & Skuladottir, 2005). Importantly, parents who are stressed may be more likely to respond to their children in ways that are not conducive to promoting quality sleep (e.g., not encouraging self-settling; Johnson & McMahon, 2008). Thus, for parents of children with CF, the stress related to managing their child's illness may contribute to poor promotion of sleep conducive behaviours, and consequently lead to further sleep problems and associated impacts (Sivertsen et al., 2009). Moreover, increased levels of parental stress have been associated with poor parental sleep (McQuillan et al., 2019), and it has been suggested that parental sleep quality may mediate the relationship between a child's illness and parents' levels of anxiety, depression and fatigue (Meltzer & Mindell, 2006). Thus the relationship between parental wellbeing and a child with CF's sleep is complex, as various mechanisms may come into play.

Furthermore, many studies have identified that despite the presence of a chronic illness, parents sleep is negatively impacted when attending to a child's NW (National Sleep foundation, 2004, Byars et al., 2020). Specifically in regard to CF, a study by Byars et al. (2020), examined the rates and impact of sleep problems among children with CF (M age = 8.8 years, $n = 91$) and their parents ($n = 79$). In their study, parents completed a sleep questionnaire which investigated common sleep outcomes, as well as CF-specific items related to problems known to fragment sleep in those with CF. They found that 40.5% of parents reported concerns about their own sleep, and nearly one third had a concern for their child's sleep. It was also discovered that many parents and children were not receiving adequate sleep, and that a significant association was discovered between child and parent sleep problems, even when the child was young and relatively healthy. These findings are

important as they illustrate that parents of a child with CF may also be facing high rates of sleep difficulties which likely intensify their already significant illness-related burden.

Sleep Interventions for Children with Cystic Fibrosis

Unfortunately, sleep problems in children with CF, especially younger children and those with preserved lung functioning, are not well understood (Byars et al., 2020 & Canter et al., 2021). A better understanding of the nature of sleep problems could lead to improved clinical care, including enhanced approaches for assessment and an improved range of available treatment options (Byars et al., 2020 & Canter et al., 2021).

Although there are many interventions to improve children's sleep in the general population, there is less known about the effectiveness of sleep interventions for children with CF (Canter et al., 2021). Given the range of medical factors that contribute toward sleep problems in those with CF, the sleep intervention literature is largely focused on the use of medical treatments, such as interventions for sleep related breathing disorders and pharmacological treatments (Canter et al., 2021). However, there is emerging recognition of the need for psychosocial (including behavioural) interventions that address non-medical factors contributing to sleep problems (Canter et al., 2021), as well as approaches which consider the presence of sleep problems during periods of clinical stability (Vandeleur et al., 2017a). The section that follows will briefly discuss medical approaches to sleep treatment, followed by an explanation of psychosocial sleep interventions and their potential for use with children with CF.

Interventions for Sleep Related Breathing Disorders

The primary goal of sleep interventions that aim to improve respiratory disruptions, is to correct nocturnal hypoxemia (Ramos et al., 2013). Research, predominately in adults with CF, has demonstrated that the use of Nocturnal Supplemental Oxygen (NOD) can result in improvements in oxygen saturation during sleep (Spier et al., 1984; Milross et al., 2001;

Gozal et al., 1997). NOD involves the administration of oxygen through nasal cannula or various masks at concentrations greater than that in natural air (20.9%), with the intent of treating or preventing hypoxia (Singh et al., 2011). Although NOD appears promising for physiological symptoms which disrupt sleep, the use of NOD has not typically translated into better SE or fewer arousals for those with CF (Milross et al., 2001; Gozal et al., 1997).

Another intervention for preventing sleep-related breathing difficulties for those with CF is the use of Non-invasive Ventilation (NIV), which supports airway clearance by increasing tidal volumes and reducing breathing effort for the person (Stanford et al., 2019). As breathing becomes difficult for those with CF (due to airway obstruction, mucous plugging, bronchial inflammation and parenchymal destruction), NIV is used to provide respiratory comfort and stability (Reiter et al., 2021). Non-Invasive Positive Pressure Ventilation (NIPPV) is one type of NIV and involves the use of ventilatory assistance without an invasive artificial airway (Peñuelas et al., 2007). Ventilation is delivered via a tight-fitting mask that covers the nose or both the nose and mouth (Peñuelas et al., 2007). NIPPV has been effective in preventing the fall in ventilation that occurs from Non-REM to REM sleep, and may help to stabilise lung function and slow down disease progression (Milross et al., 2001). Studies have found that NIPPV and supplemental oxygen therapies result in similar improvements in nocturnal oxygen saturations (Milross et al., 2001; Gozal et al., 1997; Young et al., 2008). Additionally, NIPPV is reported to result in a lower frequency of respiratory events during sleep, as compared to supplemental oxygen or room air (Milross et al., 2001; Regnis et al., 1994). However, while beneficial, no improvements have been reported in TST, SE, or frequency of arousals following the use of NIPPV (Milross et al., 2001; Gozal et al., 1997; Regnis et al., 1994).

Pharmacological Approaches

Pharmacological approaches such as the use of benzodiazepines, zolpidem and melatonin have strong empirical support for the treatment of insomnia in adults, however there is little research on their effectiveness with children, and in a number of cases they may be inappropriate for children with CF (Owens et al., 2003; Pelayo & Dubit, 2008). Furthermore, while medication may often be used to treat a variety of sleep concerns, there is actually limited data regarding appropriate dosage, effectiveness, tolerance, and safety in children (Society of Clinical Child & Adolescent Psychology, 2021).

Benzodiazepines are a class of psychoactive drugs which lower brain activity and have been associated with improvements in sleep latency and duration (de Mendonca et al., 2021). However, benzodiazepines can depress respiration, meaning that they may not be appropriate for those with CF (Shakkottai et al., 2018). Zolpidem is another commonly used sleep medication, however it has been associated with an increased risk of respiratory infections (Huang et al., 2014). Therefore, it may also not be suitable for those with CF. Lastly, melatonin is commonly prescribed to improve sleep for children, regardless of the presence of a chronic illness, due to empirical evidence demonstrating reductions in SOL and increases in TST (Malow et al., 2016; Schroder et al., 2019). A randomised, double-blinded, placebo-controlled trial of the use of melatonin in children with CF found significant improvements in SE, and a trend towards improvements in SOL following the provision of melatonin (de Castro-Silva et al., 2010). While these findings are promising and there are few known side effects, some studies have questioned the long-term safety of melatonin for use in children, as it is possible melatonin may negatively affect various developing systems, including reproductive, cardiovascular, immune, and metabolic systems (Kennaway, 2015; Anderson et al., 2016).

Thus, the consensus in the literature is that pharmacological approaches should only be used after behavioural and cognitive strategies have been properly trialled and found ineffective (Jacobs et al., 2004). This recommendation is especially important for children with CF, as sleep-inducing medications may not be appropriate due to a lack of understanding about the ways in which CF medications and sleep medications interact (Merz & Tomfohr-Madsen, 2016). It is also noteworthy that children with CF may already be on various medications which have sleep disrupting side effects (Shakkottai et al., 2018), such as albuterol (used to relax the muscles in airways; Johnson et al., 2022.), trimethoprim-sulfamethoxazole (used to treat bacterial infections; Kemnic & Coleman, 2022), and prednisone (used to suppress the immune system and decrease inflammation; Puckett et al., 2022). Finally, it is important to acknowledge that the habitual use of sleep medications for these children may further disrupt their sleep cycles and contribute to a reliance on medication to fall asleep (Merz & Tomfohr-Madsen, 2016).

Assessment of Sleep Problems

Sleep problems are complex and can occur in various combinations of difficulties, thus it is important to acknowledge that there may also be various biopsychosocial and behavioural factors that contribute toward the sleep problem (McLay et al., 2022). Before an intervention for sleep difficulties can be implemented, it is therefore important that a comprehensive assessment is conducted to identify the underlying contributing factors (Owens et al., 2002). This is important to also determine that the sleep problem is a result of modifiable environmental factors, rather than being a symptom of a medical problem (Owens et al., 2002), which is especially important for children with CF.

Functional Behavioural Assessment of Sleep Problems

Functional Behaviour Assessment (FBA) is a type of assessment which utilises the principles of operant theory, where behaviours can be understood as learned and functional

(McLay et al., 2022). FBA aims to systematically identify the relationship between antecedents, behaviours and their consequences in order to understand the purpose (i.e., function) that the behaviour serves (Blampied, 2013). FBA is an important component of any sleep intervention as it can provide a conceptualisation of the factors that underly a sleep problem so that the clinician can then develop hypotheses on the behavioural functions of the problem (McLay et al., 2022). With regard to sleep problems, FBA can be used within naturally occurring settings (such as the family's home) to identify the antecedents and reinforcers that contribute to and maintain a behaviour (McLay et al., 2022). There are three assumptions which underpin the use of FBA, these are: (1) that contexts influence how behaviour is exhibited and understood; (2) that learned behaviour serves a purpose; and (3) that problem behaviours can be reduced through teaching alternative skills that serve the same or similar function as did the problem behaviour (McLay et al., 2022). The use of FBA for informing sleep interventions is essential as behaviours do not always have the same function across individuals, therefore interventions that target the factors underlining sleep problems are going to be more effective than interventions that do not (McLay et al., 2020).

Within the context of assessing a child's sleep problem, prior to conducting an FBA it is important to gather information on the history and context of any sleep problem(s), and then objectively define these problem(s) (McLay et al., 2022). This can then be followed by conducting an FBA that aims to identify controlling variables, and consequently develop hypotheses regarding the function of the sleep problem(s) (McLay et al., 2022). With FBA, sleep problems can be assessed using a variety of measures, including information from clinical interviews, home observation, parent- and self-reported sleep diaries, psychometric questionnaires and videosomnography (Blampied & Bootzin, 2013; McLay et al., 2022). This information can then be utilised within an intervention plan which addresses the family's goals, targets antecedents (i.e., bedtime routines) and consequences (i.e., reinforcers), and

that teaches replacement behaviours, monitors progress, considers safety, and identifies the maintenance of improvements (McLay et al., 2022).

A Behavioural Model of Sleep Disturbance

Behavioural theories propose that behaviour is not only observable, but can be developed and changed through learning (Meltzer & Crabtree, 2015). A behavioural model of sleep, which like FBA utilises operant behaviour theory, can help provide an understanding of why sleep problems develop, how they are maintained and what can be done to prevent or to reduce them (Blampied & France, 1993; Meltzer & Crabtree, 2015). Operant theory proposes that behaviours operate in environmental contexts, where antecedents (i.e., events that precede behaviour) and consequences (i.e., events that follow behaviours) are provided (Blampied, 2013). Antecedents signal behaviour to occur, and consequences either reinforce (strengthen) or punish (weaken) the behaviour (Skinner, 1963). This creates a three-term contingency, or 'ABC' (antecedents, behaviour, consequences) model (Skinner, 1963).

Another core component of the behavioural model refers to contingencies of reinforcement. These can be both positive and negative. Positive reinforcement increases the likelihood that a behaviour will occur by introducing a preferred stimulus after the behaviour has occurred, such as attention or access to a tangible reward (Meltzer & Crabtree, 2015). However, with negative reinforcement, the likelihood a behaviour will occur is increased by the removal of an unpreferred stimulus (Meltzer & Crabtree, 2015). For example, a child who does not like wearing their pyjamas and throws a tantrum until the parent removes their pyjamas, is negatively reinforced for their tantrum behaviour when the pyjamas are removed, and this behaviour is therefore likely to re-occur in future (Meltzer & Crabtree, 2015).

Reinforcement is often provided by parents unintentionally (Meltzer & Crabtree, 2015). For example, at bedtime a child may not wish to sleep alone and call out to sleep with their parent. Although the parent may prefer the child to sleep in their own bed, the parent

may instead let the child sleep in the parent's bed to stop the child's crying. In this example, the child's behaviour is negatively reinforced by being removed from their bed and positively reinforced through parent attention. In this situation the parent is also negatively reinforced through the cessation of their child's crying. Such interactions are then likely to continue when the behaviours are mutually reinforcing for the parent and child (Blampied & France, 1993). Therefore, in order to reduce sleep problems, parents may also need support to learn to modify their own behaviours to improve their child's sleep.

Sleep can also be understood as a primary reinforcer because it is a biobehavioural state that human beings are fundamentally driven to enter (Blampied & France, 1993). The behaviour of falling asleep is reinforced by the consequence of sleep itself (Bootzin, 1977; Blampied & France, 1993). Thus, the act of falling asleep and the conditions associated with it are learned and modifiable (Jin et al., 2013). Therefore sleep itself is not necessarily considered a behaviour, and instead it is the behaviours required to enter and maintain sleep which become the focus of behavioural sleep interventions (Blampied & van Deurs, 2021). The act of falling asleep can also be understood as an operant chain, beginning with bed-preparation behaviours (e.g., brushing teeth, putting pyjamas on, reading stories) and ending with behavioural quietude (i.e., laying calmly in bed), which precede sleep onset (Blampied & France, 1993). However, choice occurs at every step within a behavioural chain, which may alter the path toward reinforcement (Mazur & Fantino, 2014). For example, the completion of steps in the bedtime routine can experience competition from sleep-interfering behaviours (e.g., wanting to use electronic devices) that may disrupt the chain of events leading to sleep.

The operant chain of behaviours leading to sleep are also under stimulus control. Stimuli that are consistently present in the sleep environment acquire discriminative properties that signal the availability of the reinforcer, and set the occasion for sleep

(Blampied, 2013). Specific examples of common sleep cues for children include a dimly lit room with a cool temperature, and particular bedding such as pillows, blankets, and stuffed animals (Jin et al., 2013). If a parent is regularly present when a child falls asleep, then the parent's behaviours such as rocking, patting, or shushing the child, or the mere presence of the parent themself, can become discriminative stimuli for sleep (Jin et al., 2013).

It is important that children have appropriate (i.e., sleep-conducive) cues for sleep, as the conditions under which they fall asleep (e.g., where they fall asleep) affect sleep onset and maintenance (Meltzer & Crabtree, 2015). Appropriate conditions include those which the child can independently create for themselves, such as lying in bed alone to initiate sleep (i.e., self-settling; Meltzer & Crabtree, 2015). The ability to self-settle means a child is more likely to return to sleep following a NW independently, and without requiring parental assistance. Comparatively inappropriate conditions are those in which the child requires external support to initiate sleep, such as parental presence (e.g., a parent lying with a child to sleep) or parental assistance (e.g., a parent shushing a child to sleep), devices, or sleep locations other than the child's own bedroom (e.g., the couch; Meltzer & Crabtree, 2015). Under these conditions, the child pairs the skill of falling asleep with the external support that was required for sleep onset at bedtime (e.g., rocking, parental presence, bottle), and may therefore seek these conditions if they awaken in the night in order to restore sleep (Meltzer & Crabtree, 2015; Moore, 2007). For example, if when a child falls asleep their parent is present, but then is absent during the night (e.g., the parent leaves to sleep in the parent's own bed), then the parent's presence is likely to be needed for the child to re-initiate sleep if they wake. Parental presence (e.g., cuddling, feeding, laying with the child) is one of the most common predictors of frequent NW in children of all ages (Mindell et al., 2009a).

Sleep pressure (i.e., physiological motivation to sleep) increases the reinforcing value of sleep, which also increases the likelihood of the behaviours in the chain occurring

(Blampied & van Deurs, 2021). Sufficient sleep pressure is necessary for a child to fall asleep efficiently (Meltzer & Crabtree, 2015). Insufficient sleep pressure (e.g., a child is put to bed too early) may mean a child struggles to fall asleep, thereby contributing to delayed sleep onset. If children spend a long time in bed awake (e.g., spending time worrying or using technology), their bed can become associated with a state of arousal rather than with sleep, resulting in sleep onset becoming more difficult (Meltzer & Crabtree, 2015). Therefore it is important children are encouraged to have consistent and age-appropriate sleep and wake times, in order to ensure good sleep hygiene practices occur which promote optimal sleep quality and quantity (Mindell et al., 2009a).

Behavioural Sleep Interventions

Behavioural sleep interventions focus on modifying the environment and social-interactions to facilitate behaviour change. This can include a variety of techniques such as extinction, reinforcement, antecedent modifications, and targeted skill acquisition (Carnett et al., 2020; McLay et al., 2020).

Numerous studies have found evidence of the effectiveness of behavioural sleep interventions for improving sleep in healthy children. For example, Meltzer and Mindell (2014) conducted a meta-analysis investigating the use of behavioural sleep interventions for improving paediatric insomnia among children aged 0 – 17. Their findings demonstrated significant improvements in regards to SOL, the frequency and duration of NW and SE, all with small to large effect sizes. However, they did note that there is currently little evidence for the treatment of sleep problems in older children and adolescents, as well as children with neurodevelopmental disabilities, mood disorders, and/or chronic illnesses due to a lack of studies in these populations (Meltzer & Mindell, 2014). Importantly, Meltzer and Mindell (2014) also acknowledged that although the interventions were focused on supporting the child, the parent was often the person who implemented it.

In another recent review of behavioural sleep interventions, Park et al. (2022), evaluated the effectiveness of behavioural sleep interventions in improving sleep quality and maternal depression in mothers' and their healthy children (children aged 0 – 3 years). Park et al. (2022) defined behavioural sleep interventions as practices that focused on consolidating sleep and extending TST, or where a series of consistent and repetitive activities were carried out before bedtime, which were age appropriate. Ten studies were identified which all demonstrated that behavioural sleep interventions, including settling and bedtime routine interventions, significantly reduced child sleep problems as reported by parents. Additionally, mother's sleep quality also improved following interventions, however inconsistent with previous studies, maternal depression did not improve.

Despite there currently being no research assessing behavioural sleep interventions in children with CF (Canter et al., 2021), there is a very small number of studies that have looked at behavioural sleep interventions in children with other CHC. Given children with CF may suffer similar illness experiences to children with other CHC, the effectiveness of these interventions may be useful in understanding if behavioural sleep interventions will also be effective within children with CF. Therefore research investigating these interventions in children with other CHC will be explored in Chapter Two.

The below section will describe the different strategies within behavioural sleep interventions which may be effective in reducing sleep problems in children with CF, including sleep hygiene, faded bedtime, sleep restriction, extinction procedures, and sleep education.

Sleep Hygiene

Sleep hygiene, is the term given to describe modifiable parent and child sleep practices which promote healthy sleep (Mindell et al., 2009a). Inadequate sleep hygiene has been reported to be the most common sleep issue among children (Moore, 2007). Aspects of

sleep hygiene which can be targeted to improve sleep in children with CF include avoiding

high-energy play or exercise right before bed (Denlinger et al., 2014), creating a sleep

conducive environment (i.e., comfortable, dark, and quiet), maintaining regular sleep

schedule (i.e., consistent bed and wake time), maintaining a bedtime routine, avoiding sleep-

interfering activities (e.g., the use of devices in bed), and creating strong associations

between the bed and sleep (i.e., by not spending extended periods of time in bed awake; Merz

& Tomfohr-Madsen, 2016; Vandeleur et al., 2017a). Improvements in sleep hygiene are

regarded as necessary to address sleep problems, however they are insufficient on their own

and often additional interventions are required (Blampied & van Deurs, 2021).

Vandeleur et al. (2017b), found that poor sleep hygiene played a significant role in the

sleep difficulties experienced by children with CF, including the duration of daily screen time

and the use of electronic devices or TV at bedtime. Specifically, children who used TV at

bedtime had significantly less TST (376 vs 435 mins), with reduced SE (67% vs 78%, $p <$

0.01) and longer WASO (132 vs 77 min, $p < 0.01$) compared to those who didn't use a TV.

Additionally, children's average screen time per weekday was reported to be 218 minutes

(range 30–900), and 278 minutes (range 30–720) on weekends, with the duration of weekday

screen time correlating negatively with TST ($r = -0.28$, $p = 0.01$), and positively with

frequency of NW ($r = 0.23$, $p < 0.05$). These findings suggest that sleep hygiene practices are

related to sleep quality in children with CF, and may be an important area to target.

Stimulus control involves strengthening the associations between discriminative stimuli

within the bedroom environment, in order to provide cues for sleep onset, and reduce cues

that prompt behaviours that interfere with sleep (Bootzin & Stevens, 2005; Blampied &

Bootzin, 2013). For example, stimulus control may involve avoiding stimuli that are

associated with wakefulness, such as the using of technology in the bedroom. Stimulus

control techniques are necessary when a child's bed or bedroom no longer function as

discriminative stimuli (or cues) for sleep (Blampied & Bootzin 2011, Bootzin & Nicassio 1978, Bootzin et al. 2010). Thus for an individual with insomnia, the bed and bedroom may have become associated with incompatible sleep behaviours, such as using technology, eating, worrying, and becoming frustrated from being unable to fall asleep (Bootzin & Epstein, 2011). The purpose of stimulus control is to increase the consistent presence of appropriate stimuli prior to sleep onset (e.g., pyjamas), which consequently will act as cues for feelings of tiredness, and be reinforced by the act of falling asleep. Recommendations for stimulus control include only going to bed when feeling sleepy, maintaining a set bed and wake time, temporarily leaving the bed/bedroom after long periods of wakefulness to do a non-stimulating activity (e.g., to read a book), avoiding age-inappropriate napping and avoiding sleep-incompatible behaviours in the bed or bedroom (Bootzin et al., 1991; Edinger & Carney, 2008; Edinger & Means, 2005; Blake et al., 2017).

Good sleep hygiene has been found to be an important predictor of sleep quality within various populations, including typically developing adolescents (Malone, 2011), typically developing children (Allen et al., 2016) and children with asthma (Martin et al., 2017). Hall and Nethery (2019) conducted a recent systematic review investigating the impacts of sleep hygiene on children's sleep and discovered that there was an association between bedtime routines and increased sleep duration, decreased SOL, and reduced NW. Furthermore, findings demonstrated that independently falling asleep (i.e., self-soothing) significantly increased the child's sleep duration, decreased SOL, and reduced the frequency of NW. Finally, they also found evidence for an association between electronic technology use (before bed and during the night) and shorter sleep durations, in children as early as toddlerhood.

Various studies have investigated the use of sleep hygiene education. For example, Tan et al. (2012) investigated the use of a one-on-one sleep hygiene intervention for

improving sleep hygiene practices, sleep quality and daytime symptoms among 33 children (typically developing, aged 10-18 years). Significant improvements in sleep hygiene, sleep quality, daytime sleepiness and sedentary/light activity were reported across participants following the intervention. Specifically, sleep hygiene scores (measured by the Adolescent Sleep Hygiene Scale [ASHS]) increased significantly, and self-reported sleep disturbance scores (measured by the Pittsburgh Sleep Quality Index [PSQI]), self-reported daytime sleepiness scores (measured by the Pediatric daytime sleepiness scale [PDSS]), and parent-reported sleep disturbance scores (measured by the Sleep Disturbance Scale for Children [SDSC]) all decreased significantly.

For children with CF, there may be various barriers to introducing sleep hygiene recommendations, such as the restriction of the use of technology. Specifically, a study by Canter et al. (2021) completed a qualitative investigation of the sleep habits and perspectives towards sleep interventions among 11 youth with CF, and ten parents using semi-structured interviews. They found that despite the avoidance of technology before bed being critical for sleep hygiene, youth frequently used technology (e.g., televisions and phones) in the lead up to sleep. Importantly, while many of the youth used their devices in bed for entertainment reasons, some relied on their devices to provide distractions while they completed evening treatments. Moreover, it was found that some youth used technology to monitor their CF symptoms, or to communicate with parents about their symptom management overnight. For example, one participant with CFRD used his cell phone to view his blood sugar levels throughout the night, which was essential for his health regimen. Additionally for some youth devices were used throughout the night to communicate to parents if symptoms worsened and they were requiring treatments, or a later wake time. Therefore because of the value technology provides in terms of distracting children from their illness and keeping them safe

overnight, it may not always be feasible or safe to restrict the use of technology in children with CF.

Additionally, because CF is a serious illness and symptoms can become life-threatening, it is important that complaints of illness within the timeframe of bedtime are still critically evaluated (Merz & Tomfohr-Madsen, 2016). Given the important role of parents within CF management, it is also important that psychoeducation be provided to teach parents behavioural skills that promote optimal sleep routines, discourage sleep incompatible behaviours, and set appropriate boundaries around sleep, such as limiting television use in bed and the consumption of caffeine before bedtime (Meltzer & Mindell, 2014; Mindell et al., 2006; Mindell et al., 2009a).

Faded Bedtime and Sleep Restriction

If sleep problems persist (i.e. NW are still present) after the implementation of sleep hygiene practices, then sleep/wake rescheduling and/or sleep restriction procedures can be implemented. Such approaches are designed to improve sleep pressure and stimulus control for sleep, and to regulate children's circadian cycles.

Sleep pressure can be understood by the two-process model of sleep regulation (Borbély, 1982). Part one of this model relates to how all individual's are affected by a sleep/wake dependent homeostatic process, where sleep pressure builds during wakefulness, so that an individual is ready to sleep at night (Jenni & LeBourgeois, 2006). Part two of this relates to how this homeostatic process interacts with one's circadian rhythm (internal 24-hour body clock; Jenni & LeBourgeois, 2006). These two processes then interact to generate the timing of sleep onset and waking (Achermann, 2004).

As a child develops, it has been proposed that homeostatic sleep pressure begins to accumulate more slowly, meaning a child can then be able to stay awake for longer periods and not require naps (Jenni & LeBourgeois, 2006). Furthermore, when a child's bedtime does

not reflect their sleep needs (as determined by the interactions between their homeostatic process and circadian rhythm), sleep difficulties may occur (Jenni & LeBourgeois, 2006). It can then be understood that to eliminate bedtime struggles or NW, a child's bedtime could be adjusted to ensure homeostatic sleep pressure has built enough to ensure sleep intensity and sleep consolidation (Jenni & LeBourgeois, 2006).

Sleep/wake rescheduling involves implementing a consistent bedtime and waketime for everyday of the week (Owens et al., 1999). The specific bedtime is chosen to reflect the average time of sleep onset, and the wake time is determined through ensuring the child achieves age-appropriate sleep and has sufficient sleep pressure (Owens et al., 1999). Once a consistent sleep/wake schedule has been implemented, fading can then be used to adjust the times if needed, while ensuring the child still meets their sleep requirements (Owens et al., 1999). Research has found that delaying bedtimes in children can lead to increased sleepiness, shorter SOL and less NW's (Sadeh et al., 2003).

One example of a sleep restriction technique to increase sleep pressure is the faded bedtime procedure. This procedure involves delaying a child's bedtime so that it is within 15 minutes of their typical sleep onset time (Vriend et al., 2011), while maintaining an age-appropriate TST (Ohayon et al., 2017). In addition, a set wake time is maintained, and age-inappropriate daytime sleep is eliminated as appropriate, to increase an individual's sleep drive (i.e., motivation to fall asleep efficiently and maintain sleep overnight; Vriend et al., 2011). Once the child is reliably falling asleep quickly (i.e., within 15 minutes of going to bed), with little resistance, their bedtime is then systematically moved earlier in small increments (e.g., 15 minutes every three nights), until the desired bedtime is reached (Kodak & Piazza, 2008). This procedure also improves stimulus control for sleep by strengthening the bed as a discriminative stimulus for sleep by reducing time spent in bed awake (Piazza & Fisher, 1991). The goal of sleep restriction is therefore to ensure the time an individual

spends in bed matches their sleep needs in order to improve their overall sleep quality and SE (Wohlgemuth & Edinger, 2000).

Although there have been few studies investigating the effectiveness of faded bedtime procedures and sleep restriction among children, there is evidence that these procedures may improve TST. Firstly, a study by Piazza and Fisher (1991) utilised faded bedtime procedures in four developmentally delayed children with sleep disturbances (aged 3, 4, 13 and 19 years), and found improvements for all children following systematic delays in bedtimes. All children's TST decreased initially after the procedure began, however subsequently increased following intervention representing the desired outcome (Piazza & Fisher, 1991). Additionally, two of the four children's excessive daytime sleep decreased following intervention, and three of the four children experienced decreases in NW following intervention (Piazza & Fisher, 1991).

These findings have also been replicated by Ashbaugh and Peck (1998) in a typically developing 2-year-old using the faded bedtime with response cost protocol. The response cost component involved increasing sleep pressure by removing the child from bed (response cost) when they did not fall asleep within the 15-minute time frame (Ashbaugh & Peck, 1998). Following intervention, the child's sleep-wake cycles improved dramatically, and co-sleeping was eliminated (Ashbaugh & Peck, 1998). Specifically, the number of times the child was asleep during the desired wake times (as identified by their parents), and the number of times they were awake during ideal sleep times decreased from an average of 18 times over the course of a day during baseline, to an average of three times post intervention (Ashbaugh & Peck, 1998).

The effectiveness of a faded bedtime procedure has also been investigated among autistic children, where promising findings have been demonstrated. For example, in a study by Delemere and Dounavi (2018) of six autistic children (aged between 2-7), TST increased

and SOL decreased significantly following bedtime fading. While this research is promising, it is important to note that due to the very small sample sizes of these studies, it is difficult to confidently generalise findings to children with CF without further research.

Extinction Procedures

Extinction procedures involve the withdrawal or discontinuation of reinforcement for an undesired behaviour, with the hope that over time a decrease in the frequency of that behaviour will occur (McLay et al., 2021). Within extinction procedures, the reinforcement of behaviour is changed by modifying or eliminating the child's access to reinforcement, rather than preventing or attempting to change their behaviour (Carnett & McLay, 2022). Thus, the focus of extinction procedures is to withhold or withdraw reinforcement for a previously reinforced undesirable behaviour (Carnett & McLay, 2022).

In order for extinction procedures to be implemented effectively, the procedure must match the function of the specific challenging behaviour (McLay et al., 2022). For example, if a child engages in disruptive behaviours pre-bedtime to gain parental attention, and the parental response it to provide attention, it can be assumed that the child's disruptive behaviours are reinforced by the receival of parental attention. Therefore, in order to effectively decrease these behaviours, it may be necessary to implement an extinction program where parental responses are systematically eliminated (McLay et al., 2022). Thus, over time the child may learn that their behaviours no longer receive their intended consequence, and the disruptive behaviours may reduce.

There are two categories of extinction procedures which help to understand sleep difficulties: (1) extinction of behaviours maintained through positive reinforcement (i.e., providing parental attention); and (2) extinction of behaviours maintained through negative reinforcement (i.e., escape from bed; Carnett & McLay, 2022). An example of escape-maintained behaviours would be if during sleep onset a child continuously gets out of bed to

make requests (i.e., for water), which is allowed by their parents. In this situation, the child's

requests are reinforced by their escape from having to go to sleep (McLay et al., 2022). An

extinction program in this instance would therefore require the parents to provide necessary

requests (i.e., water) non-contingently, return the child to bed following any requests, and

systematically ignore further requests (McLay et al., 2022).

Importantly, extinction procedures do not simply discontinue reinforcement

immediately, and instead they are implemented systematically and with caution (Carnett &

McLay, 2022). This involves an assessment and analysis to understand the behaviour within

its context, and identify any antecedents and maintaining factors (Carnett & McLay, 2022).

Extinction procedures have received empirical support for treating bedtime resistance,

sleep onset difficulties, and NW within healthy children and children with developmental

disabilities aged from six months up to 12 years (Rickert & Johnson, 1988; France &

Hudson, 1990; Lawton et al., 1995; Didden et al., 1998; Mindell, 1999; Weiskop et al., 2001;

Didden et al., 2002; Thackeray & Richdale, 2002; France & Blampied, 2005; Weiskop et al.,

2005; Moore, 2012; Carnett et al., 2020; McLay et al., 2022).

Although there is good evidence for the use of extinction, many parents find it difficult

to implement (Merz & Tomfohr-Madsen, 2016). This is because systematic ignoring of sleep

interfering behaviours can lead to extended periods of crying, which may result in heightened

parental anxiety and distress, especially in vulnerable parents (France & Blampied, 2005).

Extinction practices may be especially hard for parents of children with health concerns as it

may be difficult to purposefully withhold parental attention and/or may not be safe to do so

(Merz & Tomfohr-Madsen, 2016). Thus it is important that special considerations for

children with CF be made. For example, extinction strategies may be best delivered when

active monitoring for illness is no longer necessary (Merz et al., 2016), as the safety of the

child in regard to their illness should be the priority.

A systematic review by Mindell et al. (2006) examined the efficacy of behavioural interventions for bedtime problems and NW in young children (aged 0-4 years). Within this review 19 studies which focused on extinction procedures were identified. Within these studies, 17 reported extinction methods to be highly effective in eliminating bedtime problems and NW, and improving sleep continuity (Mindell et al., 2006). Mindell et al. (2006)'s review also stated that there is evidence that extinction may produce faster improvements than another well-known strategy called scheduled awakenings.

Many studies have also investigated an alternative to standard extinction procedures, called graduated extinction. Graduated extinction retains the effective properties of extinction, however is easier for parents to adhere to (France & Blampied, 2005), and may be more suitable for those with CF. Within graduated extinction procedures, parental presence during sleep onset is gradually reduced rather than completely and immediately ceased (Turner & Johnson, 2013; Morgenthaler et al., 2006). In regard to treating children's sleep problems, the most commonly used form of graduated extinction involves incremental delays in parent's responses to their child (Carnett & McLay, 2021). With this method, the delay eventually becomes so long that it is essentially a withdrawal of reinforcement (Wiggs & France, 2000; Singh & Zimmerman, 2015; Carnett & McLay, 2021). For example, if a child's disruptive behaviour before bedtime is maintained by parental attention, a graduated extinction procedure would require the parents to gradually increase the time it takes for them to respond to their child, following an appropriate response delay schedule (Carnett & McLay, 2021). Alternatively, parents can decrease the amount of time they attend to their child until there is no reinforcing attention provided at all (Carnett & McLay, 2021). Thus parents are still ignoring inappropriate behaviours but the change is less abrupt and may be more bearable. The goal of graduated extinction procedures is therefore to help a child learn to self-sooth so that they can fall asleep independently.

In the systematic review by Mindell et al. (2006) investigating behavioural sleep interventions for bedtime problems and NW in infants and young children, 14 studies were identified that used graduated extinction techniques with healthy children (aged 0-4 years). Each of these studies reported positive intervention outcomes as indicated by a reduction in bedtime problems and/or NW. Thus, Mindell et al. (2006) concluded that graduated extinction, as applied to bedtime problems and NW, may be as effective as unmodified extinction.

Another study by Reid et al. (1999) also investigated the use of standard and graduated extinction procedures to treat bedtime refusal among 49 children (aged 16-48-months). They discovered that the standard extinction and graduated extinction groups did not differ in their effectiveness, with treatment gains maintained at the 2-month follow-up for both. Additionally, mother's post-treatment evaluations of their children's sleep problems indicated that following intervention, 87% of the children were now in the normal range (Child Behaviour Checklist [CBCL] Sleep Problems subscale), compared with only 15% at pre-treatment, and no treated children were significantly worse following treatment (Reid et al., 1999).

Sleep Conducive Behaviour

Teaching sleep conductive behaviour (sleep education) is an important component of any sleep intervention as it ensures that children and parents are aware of the importance of sleep. There are a range of sleep education strategies that can be taught to children and/or their parents, with many referencing the specific needs of their targeted population. For example, sleep education for children with CHC often include specific education on how sleep may be impacted by the child's CHC symptoms (Zupanec et al., 2017; Law et al., 2018; Zhou & Recklitis, 2020).

Common sleep education strategies often involve providing information to children and parents about sleep, such as the importance of sleep, sleep architecture, homeostasis and circadian rhythms (Lunsford-Avery et al., 2021). Educating children about the specific value of good sleep, such as the impacts on their health, cognition and achievement (Lunsford-Avery et al., 2021), may provide children with the ability to consider relevant consequences, instead of simply viewing sleep guidelines as parental rules that do not benefit them. In school-aged children, education focused interventions have been found to be effective in reducing daytime sleepiness, implementing earlier bedtimes, and improving sleep duration, NW, SOL and bedtime resistance (Lunsford-Avery et al., 2021). School-based sleep interventions including sleep education and sleep hygiene techniques have also been found to improve sleep among adolescents (Lunsford-Avery et al., 2021). Specifically, improvements have been documented in sleep knowledge, self-efficacy, motivations to improve sleep behaviours, sleep practices (i.e., reducing the use of technology before bedtime) and sleep health (i.e., maintaining regular bedtimes/waketimes) following sleep education (Lunsford-Avery et al., 2021).

In addition to sleep education, there are a number of relaxation strategies that young people can be taught to support their sleep. Relaxation strategies involve techniques that aim to reduce physiological and cognitive arousal around bedtime (Borkovec & Fowles, 1973; Nicassio & Bootzin, 1974). Common relaxation techniques include deep breathing exercises, meditation, progressive muscle relaxation, passive relaxation, biofeedback, autogenic training, imagery training and mindfulness techniques (Edinger & Carney, 2009; Hendricks et al., 2014). These relaxation strategies can be used in addition to other behavioural interventions such as stimulus control, to reduce physiological arousal around bedtime (Hendricks et al., 2014). Relaxation strategies can be particularly appropriate when a child suffers from anxiety around sleep and/or their illness. For example, teaching relaxation

strategies has been effective in helping eliminate night time fears within children aged between four and 12 years (Graziano & Mooney, 1980; McMenamy & Katz, 1989). Unfortunately there is very little recent research on the use of relaxation strategies alone to improve sleep within children, however there is one study which has used relaxation techniques to improve sleep in children with CHC, this study will be discussed in Chapter Two.

One study by Blake et al. (2016) conducted a Randomised Control Trial (RCT) to investigate the use of a cognitive–behavioural and mindfulness-based sleep intervention among 144 adolescents (aged 12–17 years) with high levels of anxiety and sleep difficulties. This study is useful as mindfulness is a type of relaxation strategy which involves breathing techniques and an awareness of the present moment to relax the body and mind (Simkin & Black, 2014). Within their study, Blake et al. (2016) discovered that as compared to the control group, the cognitive–behavioural and mindfulness-based sleep intervention group had significantly better scores in sleep quality, SOL, daytime sleepiness and anxiety. Furthermore, the adolescents in the cognitive–behavioural and mindfulness-based sleep intervention group reported that they found the mindfulness of the breath, sleep hygiene and sleep education techniques as the most helpful components of the program (Blake et al., 2016). Therefore, this study demonstrates that relaxation techniques and sleep education may be well accepted treatment components within behavioural sleep interventions for children, however further research is required.

Cognitive Approaches to Sleep Disturbance

While behavioural approaches apply learning principles to the understanding and modification of sleep problems, cognitive theory focusses on the role of cognitive processes (e.g., thoughts, attitudes, beliefs) and emotions in sleep problems (Owens et al., 2002). According to the cognitive model of sleep, insomnia is maintained by unhelpful beliefs,

expectations, and attributions about the nature of sleep (Morin et al., 1993; Blampied & Bootzin, 2013). These cognitions relate to the necessity to gain sleep of a particular quality and duration, and the possible negative consequences if expectations regarding sleep are not met (Morin et al., 1993; Brand et al., 2010; Blampied & Bootzin, 2013). Unhelpful cognitions can then lead to vicious cycles of anticipatory worry or anxiety about sleep (Morin et al., 1993; Blampied & Bootzin, 2013). During sleep onset as part of these cycles, a person's attention becomes fixed on monitoring their internal (e.g., body sensations) and external states (e.g., the sleep environment) for potential indicators of poor sleep (Harvery, 2002). In return, this hypervigilance can result in difficulties with sleep initiation due to excessive arousal and emotional distress (Harvey, 2002; Schwartz & Carney, 2012). Thus, when an individual is caught in such a cycle, the excessive worry may lead to further physiological arousal and high levels of distress, which make sleep onset even more unlikely (Harvey, 2002).

Additionally, an individual may engage in potentially sleep interfering safety behaviours. These safety behaviours are actions the individual takes in order to prevent further sleep disturbance, however, are often counterproductive (Harvey, 2002). For example, an individual may believe that being on their device before bed helps with sleep initiation, however this erroneous belief will only act to further impair their sleep (Harvey, 2002; Riemann et al., 2010; Schwartz & Carney, 2012).

Another example of a counterproductive safety behaviour which may interfere with homeostatic regulation, is the act of napping during the day (Schwartz & Carney, 2012). Although the individual may believe that this is an adaptive strategy for catching up on rest, it can further interfere with the individual's homeostatic mechanisms by decreasing sleep drive, and in in turn exacerbate sleep disturbance (Schwartz & Carney, 2012). Examples of sleep-related behaviours which can negatively impact an individual's homeostatic system include

sleeping in, spending too long in bed, going to bed too early, and inactivity (Schwartz &

Carney, 2012). These behaviours all prevent the accumulation of sleep drive throughout the

day, thus reducing an individual's ability to fall asleep at bedtime (Carney et al., 2006;

Edinger & Means, 2005).

As cognitions can contribute to the maintenance of sleep difficulties, cognitive

behavioural approaches to sleep problems may be effective in children with CF. For example,

in the qualitative study of adolescents with CF's perspectives on potential sleep interventions,

Canter et al. (2021) demonstrated that adolescents with CF experience various generalised

sleep concerns which may be amenable to cognitive interventions. Additionally, the

adolescents in this study reported a high degree of acceptance toward the potential use of

CBT-I within sleep interventions.

Cognitive Behavioural Therapy for Insomnia

CBT for Insomnia (CBT-I) is an adaptation of CBT and is time limited, structured,

goal oriented and sleep-focused, involving identification of the precipitating, predisposing,

and perpetuating factors involved in sleep disturbance (Wetzler & Winslow, 2006). The

primary goal of CBT-I is to modify the patterns of thinking and behaviour that may be

underlying sleep disturbance for an individual, such as irregular sleep–wake schedules, poor

sleep hygiene, delayed bedtimes, pre-sleep hyperarousal, and maladaptive sleep-related

cognitions (Blake et al., 2017). CBT-I involves behavioural techniques such as sleep

education, sleep hygiene, stimulus control, sleep restriction, and relaxation training, as well

cognitive techniques such as addressing negative thought processes and challenging

unhelpful beliefs about sleep (e.g., worry or rumination about an inability to fall asleep;

Blake et al., 2017).

Strong evidence has emerged from multiple systematic reviews and meta-analyses

suggesting that CBT-I improves sleep in adults (Koffel et al., 2015; Murtagh & Greenwood,

1995; Morin et al., 1994; Trauer et al., 2015; van Straten et al., 2018), however there is little research on the use of CBT-I across other age groups, or among those with developmental disabilities. Whilst there are currently no studies evaluating the use of CBT-I in adults or children with CF, multiple studies have assessed the use of CBT-I in children with other CHC, and these will be discussed in Chapter Two.

Like CBT, within CBT-I the cognitive ability of the individual needs to be considered, however few studies have investigated the adaptations that CBT-I may require to be suitable to younger (non-adult) populations. The first Randomised Control Trial (RCT) to provide evidence for the effectiveness of CBT-I in typically developing adolescents with insomnia (aged 12-19 years), was conducted by De Bruin et al. (2015). De Bruin et al. (2015) compared the use of CBT-I in group therapy, guided internet therapy, and a wait-list control. As part of the CBT-I group therapy, psychoeducation, stimulus control techniques, sleep restriction, and relaxation techniques were delivered to participants over six weekly sessions. Their findings suggested that CBT-I was successful in improving SE, SOL, WASO and TST with medium to large effect sizes across outcomes. Additionally, only small differences were found between the group based CBT-I and internet based CBT-I groups, with both interventions reaching comparable endpoints.

A meta-analysis by Blake et al. (2017) also reviewed the efficacy of CBT-I in healthy adolescents ($n = 357$, aged 10-19,), with self-reported symptoms of sleep disturbance (e.g., high scores on a screening questionnaire), or a diagnosis of a sleep disorder. Their results provided preliminary evidence for the efficacy of CBT-I within adolescents, as significant improvements were seen in perceived sleep quality and functional outcomes (daytime sleepiness, depression, and anxiety). Specifically, Blake et al. (2017) found that after interventions, subjective TST improved by 29.47 minutes, SOL by 21.44 minutes, SE by

5.34%, and WASO by a medium effect size. However the authors noted that due to

considerable heterogeneity between trials, further studies were needed.

A study by Paine and Gradisar (2011) also used an RCT to evaluate the effectiveness

of CBT-I compared with a waitlist control group in 42 children (7- 13 years) diagnosed with

behavioural insomnia. CBT-I sessions combined behavioural techniques (e.g., sleep

restriction) with cognitive techniques (e.g., cognitive restructuring). Results demonstrated

that compared to waitlist controls, the children who received CBT showed significant

improvements in the sleep outcomes of SOL, WASO and SE, but not in TST. Furthermore,

CBT was also associated with reductions in problematic sleep associations, as well as child-

reported anxiety, and these improvements were all maintained at both the one and six month

follow-up assessments.

In another RCT by Schlarb et al. (2018), 112 school-aged children (aged between 5-

10 years) with insomnia completed a CBT-I program named KiSS, which has been

specifically designed for children aged 5-10 and their parents. The KiSS program provided

parents with psychoeducation about sleep and sleep hygiene, sleep restriction, graduated

extinction techniques and cognitive strategies to address irrational beliefs of parents

concerning their child's sleep. Additionally, in accordance with their age, children were

educated about sleep and sleep hygiene, and learnt relaxation techniques (deep-breathing

techniques). Completing the CBT-I programme was associated with improvements in sleep

problems, including improved SOL, SE, and reduced NW, which persisted over the 3-month,

6-month and 12-month follow-ups. However similar to the study by Paine and Gradisar

(2011), the results indicated no changes in TST. The absence of change in TST in spite of

change in other sleep variables is however not necessarily problematic, as TST is not always

a good indicator of quality sleep. This is because children may actually be awake for long

durations during the night, however are allowed to sleep in later in the morning, thus still potentially obtaining adequate TST.

Therefore there is promising evidence that psychosocial sleep interventions (i.e., behaviour-based and/or psychological [including CBT-I] interventions) are effective in improving sleep problems. Specifically, there is evidence that sleep hygiene practices, stimulus control, sleep/wake rescheduling, extinction procedures, sleep education, and CBT-I are effective in improving sleep within healthy children and children with developmental disabilities (Lawton et al., 1995; Didden et al., 2002; France & Blampied, 2005; Weiskop et al., 2005; Mindell et al., 2006; Meltzer & Mindell, 2014; Blake et al., 2017; Carnett et al., 2020; Lunsford-Avery et al., 2021; McLay et al., 2020; Park et al., 2022). Additionally, there is evidence that behavioural sleep interventions, including sleep hygiene and sleep education strategies may be acceptable to children with CF (Canter et al., 2021), thus it is possible that the effectiveness of these interventions may be replicated in children with CF.

Special Considerations for Children with Cystic Fibrosis

Given the unique clinical characteristics of CF, there are special considerations to be made when deciding whether a psychosocial sleep intervention will be appropriate. It is likely that when implementing any sleep intervention within children with CF, special accommodations will be required, such as allowing time for medical treatments, acknowledging that illness exacerbations may impact the implementation of sleep interventions, acknowledging that it may not be possible to restrict access to devices in the bedroom, and being aware of how CF symptoms may impact sleep (Canter et al., 2021).

It is also important that sleep interventions account for variability among individuals, and provide interventions that meet the child's specific needs, especially during illness exacerbations (Canter et al., 2021). It may also be beneficial for education to be provided around the different physiological sleep disruptions those with CF experience in an attempt to

reduce illness related anxiety. Additionally, comorbid psychological difficulties, such as anxiety and depression may require specific therapy separately.

It is also important that clinicians understand that families may have already attempted to improve the child's sleep through various techniques. These failed attempts may therefore impact the families beliefs in regard to the effectiveness of new interventions.

Canter et al. (2021) also noted that when attempting to establish a behavioural sleep intervention for children with CF, clinicians may be required to become flexible with the child's sleep schedule. For example, it may be necessary to look outside of the evening routine and collaborate with parents and schools if the child requires additional sleep (i.e., starting school later or planned naps), particularly during illness exacerbations where it is difficult to combat inflexible barriers to high-quality sleep. Thus, clinicians need to be understanding of individual illness demands, and ensure the family does not feel any added stress to fulfil intervention obligations in times where direct CF symptoms should be prioritised.

It may also be beneficial that the whole family be involved in interventions, including parents and siblings, as it is likely that a child's sleep difficulties will impact other household members. Including parents into interventions is important as their involvement can help encourage the use of applied learning techniques in the home environment. Additionally, if the child is old enough, it may be relevant to consider the application of cognitive methods to provide support around depression and anxiety (Quittner et al., 2014).

Importantly, participants in the study by Canter et al. (2021) were generally receptive to the idea of engaging in behavioural sleep interventions and many were interested in learning and practising time/schedule management, stimulus control, and relaxation strategies. Therefore, it is likely that behavioural sleep interventions may be positively received by young people with CF.

Rationale for The Current Thesis

Previous research suggests that there are many factors underpinning sleep disturbance in those with CF that are not directly related to their symptoms (e.g., sleep hygiene issues, anxiety). Research on psychosocial sleep interventions in typically developing children have so far proved to be effective in improving various sleep difficulties (e.g. SOL, TST, NW), however, no such research has been conducted in children with CF. As children with CF experience sleep disruptions common to children without CF, it is likely that the same sleep interventions may be effective with this population. Children with CF experience various negative impacts from poor sleep (e.g., poor mental health, worsened physical health symptoms) which may be lessened following sleep interventions. Therefore, there is a pressing need for research that investigates the effectiveness of non-medical sleep interventions for children with CF.

The aim of this research is to identify whether psychosocial sleep interventions are effective in reducing sleep problems in children with CF. To achieve this, the current thesis has been divided into two studies. Study one involves a systematic review of the effectiveness and acceptability of psychosocial sleep interventions for sleep problems in children with CHC. As there is limited research investigating the effectiveness of psychosocial sleep interventions within children with respiratory conditions, including CF, it was necessary to extend the systematic review to include children with various CHC. Importantly, children with CF may share similar illness experiences to other children with different CHC, therefore it is hoped that the findings from these studies will inform potential sleep interventions for children with CF.

The following research questions were used to guide the design of Study one:

1. What is the current state of the literature examining the short- and long-term effectiveness, feasibility, and social validity of psychosocial sleep interventions for children with CHC?

2. How can these findings inform the design of future research examining the effectiveness of psychosocial sleep interventions for children with CHC, including CF?

3. How can these findings inform assessment and intervention processes used in clinical practice with children with CHC, including CF, and their parents/carers?

Study two then evaluates the effectiveness of a behavioural sleep intervention for treating sleep problems in a child with CF using an AB case study design. The intervention will be informed by the findings of study one and will explore the following questions:

1. Is a behavioural sleep intervention effective in the treatment of sleep problems in a child with CF?

2. Will any treatment effects be maintained at Short Term Follow Up (STFU)?

3. Will the selected sleep intervention be acceptable to the parents?

4. Will improving the child's sleep affect the child's daytime behaviour and HRQOL?

5. Will improving the child's sleep affect the wellbeing of their parents?

Chapter Two: Systematic Review

Given the lack of extant research into psychosocial sleep interventions for children with CF, it was necessary to review the literature on sleep interventions in children with Chronic Health Condition(s) (CHC). The purpose of the current systematic review is therefore to identify psychosocial interventions that have been implemented to treat sleep problems in children with CHC, and to appraise the evidence for such interventions. The outcomes of this review informed the interventions that were used within the case study (Chapter Three).

The systematic review discussed within this chapter (Chapter Two) has been prepared for publication and as such, this has informed the presentation of this chapter. This review was conducted by a team of researchers including myself, and reference has been made to the co-authors of the manuscript by using author initials. My role was to complete the final review of shortlisted studies to determine which ones met inclusion/exclusion criteria. I was also responsible for completing the analysis of the included studies, as well as the academic work including writing up all sections.

Abstract

CHC are long-lasting physical conditions which affect many children worldwide and involve specific medical needs and/or result in functional limitations. Research has found that sleep problems are a common clinical concern among parents of children with CHC, and can include frequent NW, difficulty falling asleep, difficulty maintaining sleep, daytime sleepiness and poor sleep quality. Insufficient or inadequate sleep has been associated with negative psychological and physical outcomes for children with CHC. Despite the notable amount of research investigating psychosocial sleep interventions for sleep problems in typically developing children and children with developmental disabilities, little research has focused on investigating the effectiveness and acceptably of psychosocial sleep interventions

in children with CHC. This review summarizes the empirical evidence from studies that have investigated the use of psychosocial sleep interventions to improve sleep among children with CHC. Studies were included in this review if they investigated the use of a psychosocial sleep intervention and included a child (aged 0 – 18 years) with a diagnosed CHC (i.e., physical condition which has lasted or is expected to last for > 3 months, has a known biological cause and/or includes somatic symptoms and cannot be cured or if effectively treated the child remains at increased risk of recurrence or requires ongoing symptom management). A systematic search identified 13 studies for inclusion. Studies were systematically summarised, and appraised according to participant characteristics, intervention components, dependent variables, research rigor and intervention effects. Studies utilised behavioural strategies, CBT-I or Motivational Based Interviewing (MBI) and were typically implemented by the child and parent in combination, the child alone or the parent alone. Twelve studies demonstrated positive treatment effects, with majority of outcomes also maintained at follow up for the six studies which utilised follow up. Additionally, treatment acceptability was generally high across the eight studies which accessed it, with participants reporting interventions were acceptable and informative. Despite only four studies meeting criteria to be considered of adequate or strong rigor, the use of psychosocial sleep interventions emerged as promising practice. This review highlights potential areas for future research.

Introduction

CHC, often described as "long-term conditions" (Te Whatu Ora New Zealand, 2023) or "noncommunicable diseases" (World Health Organisation (WHO), 2022) are a broad term used to describe ongoing, long-term health conditions that occur across the life cycle and involve specific medical needs and/or result in functional limitations (Crump et al., 2013; Bernell & Howard, 2016; Australian Institute of Health and Welfare, 2022; Centers for

Disease Control and Prevention [CDC], 2022; Te Whatu Ora, 2022). Definitions of CHC vary according to the duration of the condition, age of onset, expected survival, physical or psychological presentations, and cause (Perrin et al., 1993). For the purpose of this review CHC is used to refer to long-term (expected to last at least three months) physical health conditions with a known biological cause and/or involve somatic symptoms. Additionally, for this review the definition of CHC required that conditions could not be cured, and if it was possible to effectively treat, the individual would still be at increased risk of recurrence or require ongoing management.

There are many different CHC which can affect children, including juvenile rheumatoid arthritis, asthma, CF, cancer, diabetes, spina bifida, haemophilia, seizure disorders, neuromuscular disease, acquired immunity deficiency syndrome, and congenital heart diseases (Committee on Children With Disabilities and Committee on Psychosocial Aspects of Child and Family Health, 1993). These conditions vary widely in terms of their aetiology, course and severity across and within children (Bristow et al., 2018). Furthermore, children can experience multiple CHC simultaneously (Russel et al., 2019).

Although some CHC are rare, when the population is considered collectively, many children are affected by CHC (Perrin et al., 2007; Crump et al., 2013). Due to the lack of consensus on definitions, it is difficult to obtain prevalence statistics for CHC in children (Van Der Lee et al., 2007). In 2021 in the United States (U.S), more than 40% of children were reported to have at least one chronic illness, however this figure also included behaviour and learning difficulties (CDC; 2021). There is also no data on the overall prevalence of CHC among children in New Zealand, however limited data is available for some CHC. For example in New Zealand, during 2018/2019 13.1% of children (aged 2 – 14 years) had medicated asthma (Environmental Health Indicators New Zealand, 2020), in 2019 0.3% of young people (aged 0 – 24 years) had diagnoses of diabetes (Health Quality and Safety

Commission New Zealand, 2021), and in 2015 the point prevalence (prevalence on specific date) of irritable bowel disease, crohn's disease, ulcerative colitis, and inflammatory bowel disease unclassified among children (aged 0 – 16 years) was 21.7, 16.5, 3.3, and 1.9 per 100,000, respectively (Lopez et al., 2017). In New Zealand the prevalence of "long-term conditions" have also been reported to be rising, particularly within Māori and Pacific people whose illness onset occurs at a younger age (Manatū Hauora - Ministry of Health, 2022).

Due to improvements in access to diagnosis, advances in medical treatment and quality of care, the survival rate of children with life-threatening CHC has increased significantly since the 1970's (Perrin et al., 2007; Shaw & McCabe, 2008; Crump et al., 2013; Gatta et al., 2014; Runions et al., 2019; Ebeling et al., 2020). Such changes have differentially affected the prevalence of specific CHC, with increases in some conditions, (e.g., leukemia, CF and congenital heart diseases) owing to increased survival rates (Gortmaker & Sappenfield, 1984; Perrin et al., 2007). Further, other conditions (e.g., lead encephalopathy) have become less common due to improved prevention (Perrin et al., 2007). Additionally, dramatic increases in the incidence of some high-prevalence conditions have also led to an increase in the number of children with CHC (Perrin et al., 2007). For example, in the U.S, obesity rose from affecting approximately 5% of children in the 1970s to 18% in 2005 (Institute of Medicine, 2005; Ogden et al., 2006; Perrin et al., 2007). The prevalence of asthma in the U.S also doubled from the 1980s to 2007, resulting in almost 9% of children being affected (Akinbami & CDC, 2006; Perrin et al., 2007). The number of children affected by CHC is also likely to continue to rise given that is the prevalence of some of these disorders can be a function of increased incidence and survival rates (Van der lee et al., 2007).

Children with CHC face numerous direct (e.g., physiological) and indirect (e.g., school attendance) challenges compared to their healthy peers (Shaw & McCabe, 2008).

These co-occurring challenges can negatively impact children's daily functioning and quality

of life, while further complicating the management of their CHC and disease burden

(Helgeson & Zajdel, 2017). Children with CHC have been reported to be at an increased risk

for developing emotional and behavioural problems (Lavigne & Faier-Routman, 1992;

Bennet, 1994; Dantzer et al., 2003; LeBovidge et al., 2003; Smith et al., 2010; Reynolds &

Helgeson, 2011; Moreira et al., 2013; Moreira et al., 2015; Quittner et al., 2016;

Lewandowski et al., 2011a). Furthermore, the presence of depression in adolescents with

cancer has been found to be associated with various adverse health outcomes, such as poor

adherence to treatments and longer hospital admissions (Kondryn et al., 2011; Park &

Rosenstein, 2015; McGrady et al., 2016). Additionally, families who have children with a

CHC may experience limited resources in terms of time and finances (Zan & Schaff, 2015).

This may not only provide further challenges to families, but also restrict the time and money

available for other enjoyable or important activities (Smith & Kaye, 2012; Thomson et al.,

2016; Roddy, 2022). Children with CHC may also experience social challenges including

reduced independence, difficulties with peer and intimate relationships, social isolation, and

educational and occupational difficulties (Meijer et al., 2000; Suris et al., 2004; Layte &

McCrory, 2012). These challenges may also result in further psychological difficulties which

likely compound the already significant burden children with CHC face.

In addition to the secondary impacts experienced by children with CHC, due to the

many challenges associated with caring for a child with CHC, parents experience greater

levels of stress, higher rates of anxiety and depression, and poorer health-related quality of

life compared to parents of children without CHC (Cohn et al., 2020). This is important as

poor psychological functioning in parents can increase the risk of poor physical and

emotional health outcomes in their children with CHC (Yuksel et al., 2007; Smith & Kaye,

2012; Cohn et al., 2020). One of the most commonly reported difficulties experienced by

parents and children with CHC, which can further impact their overall wellbeing, is the increased presence of sleep problems.

Sleep Problems in Children with Chronic Health Conditions

Sleep is essential for wellbeing regardless of the presence of CHC, however the benefits of sleep may be even more pronounced in those with CHC who already face immense challenges (Lewandowski et al., 2011a). This is due to the associations between a lack of adequate sleep, and impaired mental health, quality of life and physical health (Chattu et al., 2018). Sleep problems are also commonly reported among children with CHC (Sadeh et al., 1998; Becker et al., 2004; Bursztein et al., 2006; Heng & Wirrell, 2006; Newman et al., 2006; Naqvi et al., 2008; Sivertsen et al., 2009; Lewandowski et al., 2011a; Tsipoura et al., 2018; Lang et al., 2021).

Children with various CHC experience sleep difficulties at a much higher rate than their healthy peers, such as; frequent Night Wakings (NW), difficulty falling asleep, difficulty maintaining sleep, daytime sleepiness and poor sleep quality (Stores et al., 1998; Cortesi et al., 1999; Labyak et al., 2003; Haim et al., 2004; Meltzer et al., 2005; Palermo et al., 2007; LaPlant et al., 2007; Meltzer & Moore, 2008; Tsai et al., 2008; Fagnano et al., 2009; Sivertsen et al., 2009; Boergers et al., 2010; Dean et al., 2010; Kothare & Kaleyias, 2010; Erickson et al., 2011; Lewandowski et al., 2011a; Palermo et al., 2011; Kaleyias et al., 2012; Stinson et al., 2014; Darwish & Abdel-Nabi, 2016; McCarthy et al., 2016; Badaway et al., 2017; Monzon et al., 2018; Tsipoura et al., 2018; Ji et al., 2021; Winsor et al., 2021). Currently there is little research on the prevalence rates of sleep difficulties among children with CHC. Furthermore, these prevalence estimates vary depending on the condition, however some medical conditions have been found to be associated with specific sleep difficulties. For example specific sleep problems have been reported for children with asthma and chronic pain, which are two of the most common CHC. Research has illustrated that up

to 66% of children with asthma experience decreased Total Sleep Time (TST), more frequent

NW's, and sleep-disordered breathing (Fagnano et al., 2009; Dean et al., 2010; Lewandowski

et al., 2011a). Asthma severity has also been linked to both objective and subjective sleep

difficulties, and conversely, poor sleep has been reported to predict the severity of asthma

symptoms (Lewandowski et al., 2011a). Additionally, research has also found that children

with chronic pain frequently report insomnia symptoms, shorter TST, poorer sleep quality

and more frequent NW's compared to their healthy peers (Haim et al., 2004; Meltzer et al.,

2005; Palermo et al., 2007; LaPlant et al., 2007; Tsai et al., 2008; Lewandowski et al., 2011a;

Palermo et al., 2011). For children with chronic pain, sleep difficulties are particularly

concerning as they can exacerbate pain symptoms, and have been shown to be associated

with increased depressive symptoms, worse quality of life, and increased pain-related

interference and intensity (LaPlant, et al., 2007; Lewandowski et al., 2010; Long, et al., 2008;

Neut et al., 2012; Palermo & Kiska, 2005; Palermo et al., 2008; Palermo et al., 2011; Fales et

al., 2015). Although less common than asthma and chronic pain, children with epilepsy have

also been found to report increases in frequency and durations of NW's, reduced Sleep

Efficiency (SE) and TST, daytime sleepiness, sleep onset delay, bedtime resistance, increased

stage shifts and reduced and fragmented Rapid Eye Movement (REM) sleep (Touchon et al.,

1991; Nunes et al., 2003; Kothare & Kaleyias, 2010; Larson et al., 2012; Al-Biltagi, 2014).

Furthermore, it is important to acknowledge that these sleep difficulties often do not resolve

on their own. For example a longitudinal study of insomnia symptoms in youth with chronic

pain, found that difficulties with sleep onset and sleep maintenance persisted over a one-year

period (Palermo et al., 2012). Therefore it is evident how sleep problems in children with

CHC may persist if left untreated (Fales et al., 2015; Law et al., 2018; Li et al., 2022).

  A recent study by Adavadkar et al. (2022) used data from the Coordinated Health

Care for Complex Kids (CHECK) programme to investigate the rates of sleep disorder

diagnoses in children with chronic medical conditions. In this study, 14% of the 16,609 children (aged 0-18) with chronic medical conditions, received a sleep disorder diagnosis as per International Classification of Diseases (ICD) codes. Among these children, Sleep Disordered Breathing (SDB) difficulties were the most frequently diagnosed sleep disorder across all age groups, yet highest in the 0–2-year age group, whereas an insomnia diagnosis was most prevalent in the 14–18-year age group. Although the study by Adavadkar et al. (2022) did not solely include chronic physical conditions, children with asthma ($n = 12,198$), children with other respiratory diseases ($n = 5,234$) and children who were overweight/obese and had other endocrine/metabolic disorders ($n = 3,850$), were the most prevalent groups. Children in the "overweight/metabolic disorders" group were also reported to be four times more likely to receive a diagnosis of a sleep-related movement disorder compared to the other children. Additionally, those in the "other respiratory disorders" group had higher odds of receiving a diagnosis of SDB, insomnia, and also sleep-related movement disorders compared to the rest of the cohort. Somewhat surprisingly, for children in the asthma group, the likelihood of receiving a sleep disorder diagnosis did not significantly differ from the rest of the cohort, except for nocturnal enuresis.

There are many factors which may contribute to sleep problems in children with CHC. These include medical, psychological and behavioural factors, as well as the processes associated with managing the CHC. Medical factors which place children with CHC at higher risk for sleep disturbance include underlying disease-related issues such as airway restriction, inflammation, coughing, and itching (Meltzer & Beck, 2012). For example, children with asthma, may experience worsening of symptoms at night due to physiological changes (Suratwala & Brooks, 2007; Lewandowski et al., 2011a). This can result in common nocturnal symptoms such as coughing, airway inflammation, wheezing, mucociliary

clearance, lower lung volume, and breathlessness which may impair sleep quality and in turn disturb sleep (Suratwala & Brooks, 2007; Yuksel et al., 2007; Lewandowski et al., 2011a).

Additionally, areas of the brain which are responsible for sleep (e.g., the hypothalamus) may become affected due to insults to the brain caused by epilepsy, brain tumours, stroke, or various CHC treatments, including chemotherapy or radiation therapy (Rosen et al., 2008; Boergers & Koinis-Mitchell, 2010). For children with epilepsy, nocturnal seizures may also be more common than daytime seizures, which can result in sleep disruptions that impact sleep quality and quantity on an ongoing basis (Kothare & Kaleyias, 2010).

There are also additional illness related factors that can impact sleep, including lengthy treatment regimens, frequent changes in environment owing to hospital admissions, disruptions to routine, associated pain, side-effects of medications and night-time interruptions for illness monitoring (Valrie et al., 2007; Boergers & Koinis-Mitchell, 2010; Lewandowski et al., 2011a; Canter et al., 2021). In a survey of sleep patterns in children admitted to hospital by Herbert et al. (2014), 53% of children were reported to experience poor sleep, and although not statistically significant, the children with the highest rates of poor sleep were those with chronic medical conditions. This increase in sleep problems among the children with chronic medical conditions was proposed by Herbert et al. (2014) to be due to symptom and emotional burdens, medications, and necessary observations by hospital staff overnight, which were noted as significant causes for disrupting sleep in these children.

In addition to medical factors, there is an immense amount of research into the interaction between CHC, psychological conditions and sleep difficulties. Specifically, behavioural and emotional problems in children with chronic illnesses have been linked to difficulties initiating and maintaining sleep (Hysing et al., 2009; Lewandowski et al., 2011a).

For youth with chronic pain, the presence of mood disturbances have been found to be predictive of sleep difficulties, including irregular sleep, longer sleep latency, and difficulties waking in the morning (Palermo & Kiska, 2005; Lewandowski et al., 2011a). Pavlova et al. (2017) also found in their study of 147 youth with chronic pain that poor sleep quality was associated with increased pain intensity and pain interference, and this relationship was mediated by the presence of anxiety and depressive symptoms. This relationship has also been illustrated in child survivors of cancer, where associations have been found between increased psychological distress and fatigue, daytime sleepiness and sleep problems (Mulrooney et al., 2008; Zeltzer et al., 2009; Lewandowski et al., 2011a).

Although to our knowledge no research has examined the mechanisms by which psychological difficulties lead to sleep difficulties among children with CHC, research suggests that depression can disrupt both homeostatic and circadian drives to sleep (Nutt et al., 2008). Furthermore, the negative cognitions and persistent patterns of rumination associated with depression can result in physiological arousal, which when occurring before bedtime can delay sleep onset (Chorney et al., 2008). This may also result in a negative cycle, where sleep difficulties lead to feelings of fatigue, academic and social difficulties, and an inability to regulate ones emotions (Dahl & Lewin, 2001; Chorney et al., 2008). A study by Johnson et al. (2006) investigated the direction of the association between insomnia and anxiety among a community-based sample of 1,014 adolescents (aged 13 – 16), using structured interviews based on DSM-IV diagnoses and found that anxiety disorders preceded insomnia 73% of the time. Furthermore, the presence of anxiety may result in night-time fears, which are common in preschool and school-age children (Gordon et al., 2007). Unfortunately, despite the relationship between sleep and anxiety being well established, the direction of this association cannot be confidently determined (Brown et al., 2018).

There are also many disruptions to a child with CHC daily routine, which can in turn disrupt sleep, such as intensive disease management requirements before and after sleep (i.e., lengthy nebulising treatments for children with CF in the morning and evening), as well as during the night for some conditions (i.e., night-time blood glucose monitoring for children with diabetes; Meltzer & Booster, 2016). For children with cancer there are also various factors that can disrupt sleep including the cancer itself, and the side effects of surgery, chemotherapy and radiotherapy (Walter et al., 2015). Additionally, medications may also impact sleep. For example in children with cancer the use of the highly effective antileukemic medication Dexamethasone is associated with altered sleep and fatigue (Hinds et al., 2007; Kaleyias et al., 2012; Walter et al., 2015).

It is also possible that modifiable environmental factors may contribute to the maintenance of sleep difficulties in children with CHC. In particular poor sleep hygiene such as inconsistent sleep/wake schedules, the consumption of caffeine and the use of electronics before bed can interfere with sleep (Meltzer & Booster, 2016; Lawless et al., 2020). This was demonstrated within a study by Lawless et al. (2020) which investigated sleep hygiene among 41 adolescents with persistent asthma (M age = 14) using the Adolescent Sleep Hygiene Scale (ASHS), Adolescent Sleep Wake Scale (ASWS), and the Pediatric Quality of Life (PedsQL). Lawless et al. (2020) found that adolescents in their study reported a multitude of poor sleep hygiene practices, including inconsistent sleep schedules, not solely using their bed for sleep, consuming caffeine during the evening, engaging in high energy activities before and during bedtime, and experiencing negative thoughts and emotions before bed, and such practices were associated with poor sleep quality. Numerous studies have also identified poor sleep hygiene practices among various CHC including CF (Vandeleur et al., 2016; Canter et al., 2021; Fauroux et al., 2021), diabetes (Bergner et al., 2018), cancer (Walker et al., 2010), arthritis (Yuwen et al., 2016), and chronic pain (Valrie et al., 2013).

Parental responses to sleep problems in children with CHC may also differ from that of healthy children. Research has shown that parents of children with CHC may make special accommodations (i.e., avoiding enforcing strict boundaries), and/or become overly protective (i.e., through exerting control, struggling with separation and engaging in excessive checking of their child; Rehm, 2000; Zupanec et al., 2010; Guite et al., 2011; Pinquart, 2013; Williams & McCarthy, 2014; Williams et al., 2014; McCarthy et al., 2016; Meltzer & Booster, 2016; Merz & Tomfohr-Madsen, 2016; Kim et al., 2020 Mariyana et al., 2021). These parental factors likely contribute to significant impacts on the sleep habits of the whole family (Meltzer & Booster, 2016). Although often well-intentioned, and difficult to avoid during illness exacerbations, these parenting behaviours can unfortunately reinforce sleep difficulties. In a study by McCarthy et al. (2016) it was found that compared with parents of healthy children, parents of children with acute lymphoblastic leukemia reported significantly more relaxed parenting practises related to their children's sleep, such as co-sleeping, which was significantly associated with increased sleep difficulties. These findings were also supported in another study by Kim et al. (2020) in children with cancer, where associations were found between parental accommodating behaviours (including co-sleeping and comforting activities) and children's sleep difficulties.

There are many adverse consequences to sleep disturbance for children with CHC, including significant negative effects on daytime functioning, learning, and wellbeing (Lewandowski et al., 2011a; Stinson et al., 2014; Hansen et al., 2016; Lawless et al., 2020; Winsor et al., 2021). For example, among children with asthma sleep problems have been found to be associated with higher rates of missed school and activity limitations (Daniel et al., 2012). These impacts are problematic as children with CHC already experience limitations to these domains due to the previously discussed illness-related factors. Furthermore, sleep disturbance can also contribute to decreased treatment adherence, which

increases the overall complexity of symptom management and disease burden for children

with CHC (Boergers, 2010; Lewandowski et al., 2011a).

Many of the symptomatic challenges faced by those with CHC can also be

exacerbated by sleep problems which lead to further impairments, including in immune

system functioning, endocrine functioning, inflammatory responses and metabolism (Bryant

et al., 2004; Banks & Dinges, 2007; Lewandowski et al., 2011a; Short & Banks, 2013; Medic

et al., 2022). This has been illustrated in the bidirectional relationship where sleep problems

exacerbate other physical and psychological consequences of the CHC, which can then

further disrupt sleep. For example, in those with type one and two diabetes, fluctuations in

blood glucose levels and hyperinsulinemia can result in a stress response that impairs sleep

(Barone & Menna-Barreto, 2011). This can then lead to sleepiness, a lack of physical

activity, and appetite dysregulation, all which provide barriers for maintaining optimal

metabolic control (Barone & Menna-Barreto, 2011). Thus, research has emphasised the

importance of optimizing sleep duration and quality for those with diabetes in order to

improve blood sugar control and manage the illness effectively (Knutson et al., 2006).

Furthermore, for children with epilepsy sleep problems have been associated with excessive

daytime sleepiness, poorer quality of life and worsening of seizures (Kothare & Kaleyias,

2010), and in a study by Larson et al. (2012), the severity of epilepsy was found to correlate

positively with child sleep dysfunction, parental sleep dysfunction and parental fatigue.

As discussed earlier, there is also a significant relationship between poor sleep and

mental health challenges, however there is little research on how sleep impacts psychological

functioning in children with CHC. However, in one study of adolescent and young adult

cancer survivors and healthy controls, after controlling for baseline depression, poor sleep,

and fatigue were found to significantly predict depression in the adolescents cancer survivors,

but not the healthy controls (Daniel et al., 2016). This study is important as it illustrates that

improving sleep may contribute to improvements in psychological functioning for children with CHC.

It is also unsurprising that when children do not sleep, neither do their parents. Parents of children with CHC experience regular sleep disturbances due to disruptions in their children's sleep, as well as the requirements of monitoring their child's illness overnight (Meltzer & Booster, 2016). Such disruption can therefore impact parents sleep quality and further compound the mental health and wellbeing challenges experienced by parents (Meltzer & Moore, 2008). In parents of children without CHC, children's sleep problems have been found to significantly predict parental sleep quality, mood, stress and fatigue (Meltzer & Mindell, 2007; Varma et al., 2020). Additionally in children with cerebral palsy, who frequently require parental assistance during the night (Hemmingsson et al., 2008), sleep difficulties have been found to be associated with poorer sleep quality in parents (Lang et al., 2021). Thus it is important to treat sleep problems in children with CHC, as resolving their sleep problems will not only disrupt the bidirectional relationship between sleep and physical health (Lewandowski et al., 2011a), but also prevent further wellbeing disruptions to the child and their family. Given the high prevalence and direct and indirect impacts of sleep problems in children with CHC it is therefore critical that we identify evidence-based approaches to treat sleep difficulties among children with CHC.

Treatment of Sleep Problems in Children with Chronic Health Conditions

Common approaches for treating sleep problems in typically developing children include psychosocial interventions (including behavioural), medications and medical interventions. For children with CHC the majority of sleep treatments are biomedical in spite of best practice guidelines recommending psychosocial approaches be prioritised. Common biomedical approaches for sleep disturbance in children with CHC include pharmacological, surgical procedures such as adenotonsillectomy, and non-invasive ventilation devices such as

CPAP (Regnis et al., 1994; Gupta et al., 2005; Marshall et al., 2009; Lewandowski et al.,

2011a; Marcus et al., 2012; Kaleyias et al., 2012; Valrie et al., 2013; Felt & Chervin, 2014;

Walter et al., 2015; Sulistyawati et al., 2021; Li et al., 2022). Importantly, treatment goals

often involve a combination of approaches and include the use biomedical treatments as well

as the promotion of healthy sleep habits and prevention strategies (Lewandowski et al.,

2011a).

Pharmacological interventions have been used to treat sleep difficulties in children

with CHC, however there is shortage of evidence on their effectiveness or safety in this

population (Lewandowski et al., 2011a). In healthy children various medications including

antihistamines, melatonin, clonidine, guanfacine, benzodiazepines, chloral hydrate and

trazodone have been used to treat sleep difficulties despite the mixed consensus on their

efficacy within children (Owens et al., 2010; Cummings, 2012; Felt & Chervin, 2014;

Espositio et al., 2019; Boafo et al., 2020; Abramova et al., 2020). Specifically there is

insufficient information regarding the tolerance and safety of these medications in children

(Boafo et al., 2020), and for children with CHC their use may be further complicated due to

the presence of other illness related medications.

Melatonin appears to be the most widely used sleep medication for children and has

been reported to be effective in children without CHC, including children with

neurodevelopmental disorders (Jan & O'Donnell, 1996; Smits et al., 2003; Van der Heijden

et al., 2007; Wasdell et al., 2007; Hoebert et al., 2009; Gitto et al., 2010; Sánchez-Barceló et

al., 2011). Melatonin has also been found to improve sleep in clinically stable children and

adults with CF (de Castro-Silva et al., 2010), however it has been suggested that through

increasing REM sleep and encouraging hypoventilation, its use could worsen nocturnal

hypoxemia (Brussels & Watson, 2015). There has also been mixed findings for the use of

melatonin in children with other CHC, with some studies noting its potential to exacerbate

symptoms within children with rheumatoid arthritis, epilepsy and asthma symptoms
(Sutherland et al., 2003; Armour & Paton, 2004; El-Awady et al., 2007). Furthermore, in
children with cancer its use is not supported (Glaser et al., 2018). However other studies have
reported that it can improve sleep and does not impact symptoms in children with epilepsy or
atopic dermatitis (Gupta et al., 2003; Elkhayat et al., 2010; Sánchez-Barceló et al., 2011; Jain
et al., 2015; Chang et al., 2016; Chang & Chiang, 2016; Abramova et al., 2020).

Non-invasive ventilation devices are also used to treat sleep problems among children
with CHC. These devices provide ventilatory support without endotracheal tubes (tubes
placed into the windpipe through the nose or mouth; Nicolini et al., 2014). Sleep can
aggravate the respiratory difficulties some children with CHC experience, thus these devices
help to improve respiration during sleep (González et al., 2002). Examples of non-invasive
ventilation devices commonly used to treat Obstructive Sleep Apnoea (OSA) include
Continuous Positive Airway Pressure (CPAP) or Bi-level Positive Airway Pressures (BiPAP;
Lewandowski et al., 2011a). CPAP is the most widely used non-invasive ventilation device
and involves the delivery of a constant level of positive pressure to open the airway
(Lewandowski et al., 2011a; Nicolini et al., 2014). Conversely, BiPAP devices deliver a
higher pressure of air during inspiration and a lower pressure of air during expiration
(Nicolini et al., 2014). CPAP has been found to be effective among children with sickle cell
disease and young adults with CF (Regnis et al., 1994; Marshall et al., 2009; Lewandowski et
al., 2011a), and BiPAP has been reported to improve the respiratory symptoms of children
experiencing severe asthma exacerbations (Kang et al., 2020).

Although targeting the physiological basis of a child's sleep difficulties can be
beneficial (e.g., targeting breathing difficulties that are causing sleep disruptions), relying on
medical or pharmacological interventions does not address the psychosocial factors that
precipitate, perpetuate, and maintain sleep problems in children with CHC, and may only

offer a partial solution. Psychosocial sleep intervention (e.g., psychoeducation, stimulus control, extinction techniques, sleep/wake scheduling) offer an alternative to pharmacological approaches and are widely recommended as the first line of treatment for childhood sleep problems in typically developing children, children with developmental disabilities and children with CHC (Mindell et al., 2006; Moturi et al., 2010; Vriend et al., 2011; Felt & Chervin, 2014; American Medical Association, 2020; Phillips et al., 2020; Society of Clinical Child & Adolescent Psychology, 2021). Various psychosocial sleep interventions have been reported to effectively reduce sleep problems (including sleep onset and maintenance difficulties) among typically developing children and children with neurodevelopmental disabilities (Mindell, 1999; Owens et al., 1999; Wiggs & France, 2000; Mindell et al., 2006; Meltzer & Mindell, 2014; Rigney et al., 2018; Carnett et al., 2020; Pattison et al., 2020; Lunsford-Avery et al., 2021).

There are currently no reviews which have addressed the effectiveness of psychosocial sleep interventions for sleep problems in children with CHC, despite psychosocial sleep interventions being considered best practice. Therefore, there is a critical need to identify empirical evidence pertaining to the use of psychosocial sleep interventions for children with CHC who experience distinct physiological and psychological challenges. Overall this review has three aims: (1) identify and appraise the literature examining the effectiveness, feasibility and social validity of psychosocial sleep interventions for children with CHC; (2) identify how these findings can inform the design of future research examining the effectiveness of psychosocial sleep interventions for children with CHC, including CF; and (3) identify how these findings can inform assessment and intervention processes used in clinical practice with children with CHC, including CF, and their parents/carers.

Methodology

This systematic review was registered with the Prospective Register of Systematic Reviews (PROSPERO), and conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Moher et al. 2009).

Search Procedure

A systematic search was undertaken using the electronic databases PsycINFO, Medline, Embase, Education Source, Cochran Library and Scopus. The initial search was conducted in July 2022 with support from a specialist subject librarian with expertise in database searching for systematic reviews. Search terms included keywords related to CHC (e.g., chronic illness, chronic disease, lifelong condition) specific diagnoses (e.g., diabetes, eczema, leukaemia); sleep (e.g., circadian rhythm, insomnia, parasomnia), treatment (e.g., psychological intervention; psychoeducation; sleep hygiene); and age range (e.g., child, adolescent, young person). Refer to Table 1 for a list of subject headings, search terms, and the number of papers produced from each database.

Inclusion and Exclusion Criteria

This review focused on specific CHC (see Table 1) that were identified in advance of the systematic search according to the following criteria: (a) were of a known biological cause and/or included somatic symptoms; (b) were chronic (i.e., have lasted or are expected to last for > 3 months, Crump et al., 2013); (c) cannot be cured, or if effectively treated the individual is at increased risk of recurrence or requires ongoing symptom management; and (d) are associated with sleep problems, as identified in the research literature. Additionally, if a study was identified which included a CHC not previously listed in the inclusion criteria, these studies were still eligible for inclusion. The CHC identified for inclusion were developed and reviewed by the wider research team.

Based on the definition provided by England et al. (2015), psychosocial interventions were considered those that "capitalize on psychological or social actions to produce change in psychological, social, biological, and/or functional outcomes" (p. 10). In regard to sleep, psychosocial interventions include behavioural (i.e., based on the principles of applied behaviour analysis) and/or cognitive techniques. Specific therapies include psychoeducation (e.g., sleep hygiene), common behavioural sleep intervention methods (e.g., extinction, reinforcement, sleep/wake scheduling) and Cognitive Behavioural Therapy for Insomnia (CBT-1; see Åslund et al., 2018 for a review). For the purpose of this review, studies which focused on children with neurodevelopmental disorders were excluded. For example, if within a study all children had neurodevelopmental disorders, the study was excluded, however if a study only included some children with neurodevelopmental disorders, and some with CHC, the study was included. This exclusion criteria was utilised as the primary aim of the review was to evaluate the effectiveness of sleep interventions for children with chronic physical health conditions. Further, children with developmental disabilities may have different underpinning etiologies of sleep problems to those of people with CHC alone. As such, response to psychosocial sleep interventions among these group could be expected to differ.

Included articles met the following criteria: (a) were published in an academic, peer-reviewed, English language journal; (b) examined the effectiveness of psychosocial interventions for sleep problems; (c) reported quantitative sleep outcome data for at least one participant aged 2-18 years with a CHC; (d) intervention primarily focused on the treatment of one or more types of sleep problems; (e) sleep interventions were implemented in the home setting, though clinical support (e.g., parent psychoeducation) may have been delivered in community or outpatient settings; and (f) interventions were delivered in-person, or via an online/digital platform, or a combination of the two. Studies were excluded if the sleep

treatment procedures were not psychosocial (e.g., pharmacological treatments or medical procedures), if all participants had neurodevelopmental disorders, and/or if the interventions were administered in an in-patient setting. Given the limited available research, no limit was placed on the date of publication or research design.

Table 1

Summary of Databases and Search Terms

Database	Search terms				Number of articles produced
	Group 1	Group 2	Group 3	Group 4	
All database	child* OR adolesc* OR teen* OR youth* OR toddler* OR school age* OR pediatric* OR paediatric* OR juvenile OR young* adult* OR young* person OR girl* OR boy* OR preadolesc* OR pubescen* OR prepubescen* OR puberty OR prepuberty OR teen*	Psychological intervention* or psychological treatment* or behav* treatment* or behav* intervention* or Therap* or psychoeducation or sleep hygiene or extinction or modified extinction or faded bedtime or sleep restriction or reinforcement or reward* or cognitive behav* therapy or CBT or "Acceptance and Commitment Therap*" or Mindfulness or relaxation or progressive muscle relaxation or visual imagery "Behavior Modification" "Applied Behavior Analysis" "Behavior Therapy" "Contingency Management" "Fading (Conditioning)" "Cognitive Behavior Therapy" "Acceptance and Commitment Therapy" "Cognitive Techniques" "Cognitive Therapy" "Early Intervention" "Family Intervention" "Group Intervention" "Mindfulness-Based Interventions" "Multimodal Treatment Approach"	Cystic Fibrosis or bronchiectasis or bronchiolitis obliterans or interstitial lung disease or Atopic Disease* or Asthma or Eczema or atopic dermatitis or psoriasis or Allergic Rhinitis or Diabetes or diabetes mellitus or type 1 diabetes or Juvenile Arthritis or rheumatoid arthriti* or ankylosing spondylitis or connective tissue disorder* or vasculitis or Irritable Bowel syndrome or Gastrointestinal disorder* or Crohn* disease* or Ulcerative colitis or inflammatory bowel disease or enteral feeding or irritable bowel syndrome or Cancer* or Haematological malignan* or tumour* or neoplasm* or paed* oncology or leukaemia or lymphoma* or neuroblastoma or osteosarcoma or migraine* or headache* or migraine or chronic pain or chronic illness* or chronic disease* or lifelong illness* or lifelong disease* or lifelong condition* or Seizure* or Epilep* or sickle cell disease or allergic rhinitis or Obes* or overweight or Congenital heart disease or Kidney Transplant or Liver transplant or Dental Health or oral health	Sleep* OR circadian OR insomnia* OR parasomnia* OR dyssomnia* OR hypersomnia* "Insomnia" "Sleep" OR "Sleep Wake Disorders" Sleep/ or sleep deprivation/ or sleep hygiene/ or sleep quality/ or sleep latency	9860
PsycINFO	As reported in all databases	As reported in all databases	As reported in all databases	As reported in all databases	1755
MEDLINE	As reported in all databases	As reported in all databases minus Cognitive Techniques" "Cognitive Therapy" "Early Intervention" "Family Intervention" "Group Intervention" "Mindfulness-Based Interventions" "Multimodal Treatment Approach" plus meditation/ or sleep phase chronotherapy/ or psychosocial intervention *Conditioning, Classical/ or Conditioning, Operant	As reported in all databases	As reported in all databases	1747
EMBASE	As reported in all databases	As reported in all databases	As reported in all databases	As reported in all databases	2515
Education Source	As reported in all databases	Psychological intervention* or psychological treatment* or behav* treatment* or behav* intervention* or Therap* or psychoeducation or sleep hygiene or extinction or modified extinction or faded bedtime or sleep restriction or reinforcement or reward* or cognitive behav* therapy or CBT or "Acceptance and Commitment Therap*" or Mindfulness or relaxation or progressive muscle relaxation or visual imagery)		As reported in all databases	169
Cochran Library	adolescen* or boy* or child* or girl* or juvenil* or paediatric* or peadiatric* or pediatric* or preschool* or teen* or toddler* or youth*	"Psychological intervention*" or "psychological treatment*" or "behav* treatment*" or "behav* intervention*" or Therap* or psychoeducation or "sleep hygiene" or extinction or "modified extinction" or "faded bedtime" or "sleep restriction" or "reinforcement or reward*" or "cognitive behav* therapy" or CBT or "Acceptance and Commitment Therap*" or Mindfulness or relaxation or "progressive muscle relaxation" or "visual imagery"	As reported in all databases	Sleep or circadian rhythm or insomnia or parasomnia or dyssomnia or hypersomnia	35 Cochrane Reviews and 1305 Trials
Scopus	As reported in the Cochran Library	As reported in the Cochran Library	As reported in all databases	As reported in all databases	2334

Figure 1 summarises the systematic search procedures for this review. Initially, 9,869 articles were identified. Once duplicates were removed, 8,245 remained. The titles and abstracts of each of these manuscripts was then reviewed. An inter-rater agreement check was first conducted in which two members of the research team independently screened the titles and abstracts of the same 100 articles. Articles for inclusion and exclusion were then compared. This process resulted in 100% agreement across reviewers. The titles and abstracts of all remaining articles was then screened against the inclusion criteria. The majority of these studies ($n = 8,218$) were excluded because: (a) sleep problems were not a target of intervention; (b) interventions for sleep problems were not psychosocial (e.g., treatment was pharmacological); (c) all participants were over 18 years of age; (d) no participants met criteria for CHC; and/or (e) all participants were diagnosed with a neurodevelopmental disorder. The remaining 27 articles were independently reviewed in full by the thesis author to determine eligibility. A total of 14 articles representing 13 research studies (Palermo et al., 2016 and Palermo et al., 2017 presented acceptability and effectiveness results across two articles) were identified for data extraction. There was 100% agreement across reviewers on the final selection of articles.

Data Extraction

Each included article was summarised (see Table 2) according to: (a) participant characteristics (number, age, gender, diagnoses); (b) the types of sleep problem(s) targeted during intervention; (c) research design and follow-up assessment procedures; (d) characteristics of the intervention approach (e.g., intervention components; dosage; agent); (e) dependent variables (including social validity, treatment fidelity) and data collection procedures; (f) intervention outcomes (e.g., effect size estimates as appropriate); and (g) overall study quality (I.e., strong, adequate, weak; see Evaluation of Study Quality below).

Figure 1

Flowchart of Search Procedures and Included Studies

Identification of Studies Via Databases and Registers

Identification

Articles returned from online databases

(n= 9,869)

→

Articles removed before screening: Duplicate articles removed

(n=1,624)

Screening

Articles assessed for relevance based on title and/abstract

(n= 8,245)

→

Articles excluded:

(n=8,218)

- Participants age range was outside 2-18 years
- Treatment focused on medical procedures or medications
- In-patient interventions
- No CHC
- Did not examine the effectiveness of psychological interventions for sleep problems

Remaining articles sought for retrieval

(n=27)

Text of remaining articles examined

(n=27)

→

Articles excluded:

(n= 13)

- Sleep interventions weren't the focus of the intervention
- Articles were not fully published
- Articles could not be located
- Health conditions were not chronic
- Quantitative data was not reported
- Was not published in an academic, peer-reviewed, English language journal

Articles accepted based on review criteria

(n=14)

Included

Final articles accepted and reviewed

(n=14)

Table 2

Summary of Included Research

Author/s	Participant Characteristics	Target Behaviours	Study Design, Follow-up	Intervention Characteristics	Outcome Measures	Results	Treatment Fidelity & Social Validity	Quality Rating
Bartlet & Beaumont, 1998	$N = 61$, 11 mnths-17y ($M = 4$y, 8 mnths), 40M:21F, 39 Ps with developmental disorders & 22 chronically ill Ps (incl., chronic upper-respiratory tract infections, eczema, deafness, asthma, epilepsy, coeliac disease). *Country:* U.K.	Settling & NW's.	Nonconcurrent multiple baseline across Ps design. *Follow up*: 3-6 mnths post-int.	*Agent:* Parent & child. *Int. Type:* Individualised behavioural strategies. Cueing (pre-bedtime routine), graded change (breaking down the sequence of change), Ext. & Rx. *Dosage:* Av. of 3 mnths. *Clinical Support:* 1.5-2hr Ax. (some required 2nd Ax.), followed by regular ph calls ($M = 4.95$ calls, varying from 5-60mins).	*Sleep Measures:* Southampton Sleep Management Schedule, SDI (pre- & post-int.), sleep diaries. *Other Measures:* GHQ-30 (measuring parent health, pre- & post-int & f/up).	*F/Up:* 45 families reported improv., 10 reported no change, & 2 reported ↓. Ps in the "disabled" & "chronic illness" categories had similar rates of improv. *SDI:* Sign. ↓ in Av. SDI scores for group (one-sample t-test: $M = 3.544$, SD 3.57, $p = 0.000$).	*Treatment Fidelity:* N/A. *Social Validity:* 27 families reported specialist help was useful & should be more readily available. 7 families reported programme difficult to manage or ineffective.	Weak
Kellerman, 1979	$N = 1$ (3 y) child with acute lymphocytic leukemia currently in remission, & experiencing persistent, recurring night terrors (1-6 times a night). *County:* U.S.	Night terrors.	Single case study. *Follow up:* Regular reporting during 246 days of int.	*Agent:* Parent & child. *Int. Type:* Mother provided with psychoeducation on separation anxiety, & trained in progressive muscle relaxation to reduce her own anxiety. Parents encouraged to provide positive rx. for nights when no night terrors occurred & use minimal reassurance when they did occur. Child received psychotherapeutic sessions to desensitize to medical apparatus & ↑ tolerance to separation from mother. *Dosage:* 246 days. *Clinical Support*: Sessions were gradually ↓ to weekly, bi-monthly & monthly schedules	*Sleep Measures:* Daily night terror logs (parent report). *Other Measures*: Nurse & researcher feedback.	*Night terror logs:* Over 246 days the child's night terrors ↓ & became symptom free for 120 days. She also had no night terrors following a 4th BMA. *Nurse & researcher feedback:* Post-int, the mother was able to remain with child during BMA's (unlike pre-int.), which helped with BMA's. The child's separation anxiety ↓ post-int.	*Treatment Fidelity:* N/A. *Social Validity:* N/A.	Weak
Law et al., 2018	$N = 21$, 11–17y ($M = 15.5$y), 4M: 17F, with Dx. of chronic migraine & experiencing insomnia. 81% Anglo American. *Country:* U.S.	Headache freq. & insomnia symptoms.	Single-arm pilot trial (pre-post-test). *Follow up:* 3 mnths post-int.	*Agent:* Adolescents & parents. *Int. Type:* CBT-I, incl. sleep hygiene & headache psychoeducation, stimulus control, sleep restriction, relaxation training, pleasant activity scheduling, positive thought tracking, anxiety & fatigue management. Parents received operant training to Rx adolescent skills practice. Adolescents completed weekly homework. *Dosage:* 6-12 weeks. *Clinical Support:* 6 individual sessions (in person, each lasting 60-	*Sleep Measures:* Sleep diaries (adolescent report for 7 days at pre- & post-int. & f/up), ISI, ASWS, & ASHS (all adolescent report, at pre-, post-int. & f/up). *Other Measures:* Headache diary (adolescent report for 7 days at pre-, post-int. & f/up), CALI-21	*Sleep Diaries:* Post-int. adolescents reported: sign. ↑ in SE ($b = 9.31$, $p = .008$, $d = −.60$) & f/up ($b = 13.51$, $p = .001$, $d = −.95$), sign. ↓ in WASO & SOL from pre- to post-int. ($b = −22.98$, $P = .012$, $d = .73$; $b = −38.28$, $p = .015$, $d = .71$ respectively) & f/up ($b = −23.37$, $p = .01$, $d = .74$; $b = −41.87$, 15.21, $d = .67$ respectively). TST was stable from pre- to post-int. ($b = 7.53$, $p = .776$, $d = −.02$) & ↑ sign. from pre-int. to f/up ($b = 89.85$, $p = .003$, $d = −.56$). *Sleep Outcomes (ISI, ASWS, ASHS):* Post-int. adolescents reported sign. ↓ in insomnia symptoms ($b = −7.32$, $p = .001$, $d = 1.31$), & f/up ($b = −7.60$, $p = .001$, $d = .50$). Sleep quality & sleep hygiene sign. ↑ from pre- to post-int. ($b = .74$, $P = .001$, $d = −1.32$; $b = .51$, $p = .001$, $d = $	*Treatment Fidelity:* Monitored in weekly supervision using case conference format. Therapists rated Ps as highly compliant with homework completion ($M = 9.50/10$, SD = .74). On Av., Ps completed 5/7 diary days at each Ax. point.	Weak

				90 mins), & a booster session 1 mth post-int.	(adolescent report, at pre-,post-int. & f/up), TEI-SF (parent & adolescent report, post-int.).	−1.09, respectively), & f/up (b = .67, p = .002, d = −1.06; b = .42, p = .008, d = −.73, respectively). **Headache Diary:** Post-int adolescents reported sign. ↓ in headache freq. (b = −1.91, P = .004, d = .84), maintained at f/up (b = −2.16, p = .002, d = .87). Headache pain intensity unchanged from pre- to post-int. (b = .40, p = .25, d = −.28) or f/up (b = −.15, p = .68, d = −.28). **Exploratory Analysis:** ↓ in headache freq. from pre-int. to f/up strongly correlated with ↓ in insomnia symptoms from pre-int. to f/up (r = 0.50). **CALI-21:** Activity limitations stable from pre- to post-int., & scores sign. ↓ at f/up (b = −11.57, p = .029, d = .69).	**Social Validity:** TEI-SF – Av. parent score = 40.67, SD = 4.48; Av. adolescent score = 39.13, SD = 5.10, indicating "moderate" acceptability. Of the 21 enrolled Ps, 4 did not complete int. (due to a major health event). Remaining Ps completed all sessions.	
Meltzer & Booster, 2016	N = 89 (Dx. of atopic dermatitis & asthma), 89% "white" ethnicity. Int. (N = 53), (M = 6.96y), 19M:34F. CG (N = 36), (M = 7.47 y), 17M:19F. No sign. diff. between groups. *Country*: U.S.	Sleep habits & sleep quality.	Non randomised controlled study. (Int. vs CG). *Follow up:* N/A.	**Agent:** Parents. **Int. Type:** Int. group received a 1-hr sleep health group int. (providing sleep health education & strategies to ↑ sleep). **Dosage:** 1-hr. **Clinical Support:** Not spec.	**Sleep Measures:** CRSP (parent report of child's sleep), PSQI, ISI & SHI (parents sleep). All completed pre- & post- int. **Other Measures:** N/A.	**CRSP:** Results revealed a sign. time effect for the activities in the hr before bed index, F(1, 86) = 70.64, p < .001; the sleep location index, F(1, 86) = 12.45, p < .001; the use of electronics at sleep onset index, F(1, 86) = 19.27, p < .001; & the insomnia scale, F(1, 82) = 15.20, p < .001. However, no effect for group was found. **PSQI:** Results revealed a sign. effect for time, F(1, 84) = 29.92, p < .001, but no sign. interaction between time & group (i.e. parents reported ↑ in their sleep quality 1 mnth post-discharge, regardless of group). **ISI & SHI:** No sign. effects for time or group.	**Treatment Fidelity:** N/A. **Social Validity:** N/A.	Weak
Palermo et al., 2016, 2017	N = 40, 11-18y (M = 14.9), 10M:30F, 85% Caucasian, all with co-occurring medical & mental health conditions (Conditions: 10 Ps: physical, 11 Ps pscyh, 19 Ps physical & pscyh) & insomnia. *Country*: U.S.	Consistent sleep/wake schedule.	Single-arm pilot trial (pre-post-test). *Follow up:* 3 mnths post-int.	**Agent:** Adolescent & parent. **Int. Type:** CBT-I involving sleep education, sleep hygiene, stimulus control & sleep restriction. Optional: addressing bedtime resistance & co-sleeping, anxiety, relaxation strategies, cognitive restructuring, social support for sleeping away from home & parental guidance on positive rx., Modules incl. handouts & worksheets. **Dosage:** 4-6 weeks. **Clinical Support:** Up to 4 sessions (max 75mins) in person delivered individually or conjointly with adolescent & parent.	**Sleep Measures:** Sleep diaries & actigraphy (adolescent report for 7 days at pre- & post-int. & f/up), ISI, ASWS-R, ASHS, & PSAS (all adolescent report at pre- & post-int & f/up). **Other Measures:** PROMIS Pediatric Anxiety Short Form & Pediatric Depressive Symptoms Short Form, & PedsQL (all adolescent report at pre- & post-int. & f/up), therapist engagement progress note (post- sessions), TEI-SF (parent & adolescent post-int.).	*Reported by Palermo et al., 2017:* **ISI:** Sign. ↓ from pre- to post-int. (b = −5.67, p < 0.0001; M pre = 15.10, SD = 4.34; M post = 9.96, SD = 4.96; d = 1.23), sustained at f/up (b = −5.69, p < 0.0001; M f/up = 9.67, SD = 5.98; d = 1.13), representing a large magnitude change. **ASWS-R:** Sign. improv. with med - large ES, (b = 0.74, p < 0.0001; total M pre = 3.23,SD = 0.69; M post = 3.99, SD = 0.77; d = −1.03; M f/up = 3.96, SD = 0.84; d = −0.92). **ASHS:** Sign. improv. with med - large ES, (b = 0.43, p < 0.0001; M pre = 4.65, SD = 0.59; M post = 5.11, SD = 0.53; d=−0.81;b=0.38,p<0.0001; M f/up = 5.05, SD = 0.55; d = −0.70). **PSAS:** Sign. ↓ from pre-to post-int. (b = −6.97, p < 0.0001; M pre = 39.50, SD = 12.73; M post = 31.97, SD = 12.32; d = 0.60), (med ES), sustained at f/up, (M = 30.56, SD = 10.98; d = 0.73). **Sleep diaries:** Small ES from pre- to post-int. in SOL (b = −33.03, p < 0.03; SOL M pre = 92.4min, SD = 82.7min; M post = 59.0min, SD = 104.6min; d = 0.35) & SE (b = 5.65, p = 0.02; SE M pre = 82.4%, SD = 14.1; M post = 88.0%, SD = 17.0;d = −0.36), both maintained at f/up with med ES. Sign. ↓ in WASO from pre- to post-int. (b = −8.22, p = 0.003; WASO M pre = 15.3min, SD = 18.2min; M post = 7.2min, SD =	*Reported by Palermo et al., 2016 & Palermo et al., 2017:* **Treatment Fidelity:** Ps rated as compliant with homework completion (M = 7.5, SD = 1.7). Researcher reported Ps were generally compliant with scheduled visits. **Social Validity:** TEI-SF (completed by 38 families) - Av. parent score = 38.8, SD = 5.2; Av. adolescent score = 36.8, SD = 4.6, indicating "moderate" satisfaction. 85% of Ps completed all 4 sessions (M = 3.7, SD = 0.76).	Weak

Study	Sample	Targets	Design	Intervention	Measures	Results	Fidelity / Social Validity	Rating
						12.3min; *d* = 0.51) & from pre-int. to f/up (*M* f/up = 4.8min, SD = 6.2min; *d* = 0.70), with med ES. TST unchanged across Ax.'s. *Actigraphy:* Few changes detected. *Pediatric Anxiety Short Form:* Symptoms sign. ↓ from pre- to post-int. (*b* = −3.32, p = 0.02; *M* pre = 54.2, SD = 13.6, *M* post = 50.3, SD = 13.1; *d* = 0.29) & this small ES was maintained at f/up (*M* f/up = 49.3, SD = 12.8; *d* = 0.37). *Pediatric Depressive Symptoms Short Form:* Symptoms sign. ↓ from pre- to post-int. (*b* = −3.36, p = 0.04; *M* pre =54.4, SD =13.3, *M* post =51.2, SD =10.8; *d* =0.25) & to f/up (*b* = −3.58, p = 0.03; *M* f/up = 50.4, SD = 13.1; *d* = 0.30). *PedsQL:* No sign. change in total QoL or physical & psychosocial QoL from pre- to post-int. However, sign. ↑ from pre-int. to f/up on total HRQOL scores, reflecting (med. ES).		
Sonney et al., 2020	*N* = 25, 6-11y, (*M* = 8.3), 13M:12F, 76% Caucasian, all experiencing persistent asthma & sleep disturbance. *Country:* U.S.	Sleep quality, bedtime routine, sleep environment.	Prospective single group design (pre-post-test). *Follow up:* 12 weeks post-int (week 24).	*Agent:* Parent & child. *Int. Type:* Ps were provided with feedback from baseline actigraphy & then were recommended 1+ Sleep Intervention for Kids & Parents (SKIP, dev. by authors) online modules (focused on: bedtime routine, sleep quality &/or bedtime environment, each lasted 30mins). Weekly module specific content (incl. educational content, behavioural strategies, elements of CBT-I & goal setting) & SKIP activities were assigned. SKIP provided tips in response to Ps goals, & during weeks 2–8 of int., Ps completed progress reports. *Dosage*: 8 weeks. *Clinical Support:* No clinical support spec. after initial feedback/guidance.	*Sleep Measures:* Actigraphy & sleep diaries (accessing child & parent, for 10 days at pre-, post-int. & f/up). *Other Measures:* Online survey (individually) & semi structured interview (parent & child together) to assess int. acceptability (f/up).	*Actigraphy & Sleep diaries: Child:* Post-int, sign. ↑ for: TST (Cohens *d* = 0.31, *P* <.001, 95% CI), WASO (Cohens *d* = 0.72, *P* <.001, 95% CI), SE (Cohens *d* = 0.69, *P* <.001, 95% CI), & bedtime range (Cohens *d* = 0.37, *P* <.001, 95% CI) , but not for bedtime coefficient of variability (Cohens *d* = 0.1, 95% CI). WASO, SE & bedtime range scores maintained at f/up, but TST ↑'s not sustained & scores resembled pre-int.	*Treatment Fidelity:* N/A. *Social Validity:* 61% of children & 92% of parents reported overall satisfaction with the int.	Weak
Tsai et al., 2020	*N* = 100, 1.5–6y (*M* = 3.87 y), 55M:45F, with epilepsy (44.0% w seizures in past 3 mnths). 43% with co-occurring developmental delays. Int. (*N* = 50); Usual Care with attention (*N* = 50). *Country:* Taiwan.	SE, TST, daily sleep duration (over 24hrs).	RCT (int. vs usual care). *Follow up:* N/A.	*Agent:* Parent. *Int. Type:* Sleep, Health, & Research for Epilepsy (SHARE) program (dev. by authors). Used actigraphy data to provide parents with sleep education & strategies to promote sleep. *Dosage:* 12 mnths. *Clinical Support:* Both groups received routine clinic visits every 3 mnths. Int. group received additional 3 face-to-face sleep education sessions (session 1: 45-60mins, sessions 2 & 3 at 3 & 6	*Sleep Measures:* Actigraphy & sleep diaries (completed for 7 days at 3, 6 & 12 months), CSHQ, PSKI, & PSQI (all pre- & post-int.). *Other Measures:* N/A.	*Actigraphy:* Sign. ↑ in SE at 3- & 6- mnths (1.92% (p < .01) & 2.35% (p < .01) respectively) for int. group. SE unchanged for the usual care group. Post-int. after adjusting for demographics, int. group had > SE by 2.03% compared with the usual care group (95% CI = 0.20% to 3.86%; p = .03). Post-int., the int. group Av. TST was sign. longer; adjusted *M* diff. of 16.13mins (95% CI = 0.24% to 32.03%; p = .04). No sign. diff. between groups for daily sleep duration post-int. (95% CI = −10.63% to 26.66%; *p* = .39). *CSHQ:* No sign. diff. between groups post-int. (95% CI = -2.59% to 2.08%; *p* = .82). *PSKI:* No sign. diff. between groups post-int. (95% CI = -3.11% to 5.20%; *p* = .61).	*Treatment Fidelity:* 1st author provided monthly supervision & reviewed Ps progress. Randomly chosen subset of 10 int. sessions recorded & reviewed by first author to ensure fidelity. *Social Validity:* Researcher reported int. well accepted by families.	Strong

				mnths: 30-45 mins). Post-int. ph call at 9 mnths to review progress.		*PSQI:* No sign. diff. between groups post-int. (95% CI = -0.91% to 0.98%; *p* = .94).		
Tumakaka et al., 2019	*N* = 46, 6-18y, with Type 1 Diabetes Mellitus (T1DM). Int. group: *N* = 23. CG: *N* = 23. *Country:* Indonesia.	Sleep quality.	Pre-post quasi-experimental design, using non-equivalent CG. *Follow up:* N/A.	*Agent:* Parent & child. *Int. Type:* Int. group received 10mins of sleep hygiene education via video, & then were asked to practice sleep hygiene tips for 3 days & complete a sleep hygiene practice sheet. CG continued usual sleep habits *Dosage:* 10mins. *Clinical Support:* Not spec.	*Sleep Measures:* PSQI (parent report, pre- & post-int.). *Other Measures:* N/A.	*PSQI:* Int. group Av. score sign. improv. at pre- & post-int. (*p* < 0.001). No sign. diff. between pre- & post-int. for CG (*p* = 0.833). There was a sign. diff. between groups post-int. (*p* = 0.001).	*Treatment Fidelity:* N/A. *Social Validity:* N/A.	Weak
Wiggs & Stores, 1998	*N* = 30 children with a severe sleep problem & challenging behaviour (incl. children current uncontrolled epilepsy). Int. group: *N* = 15 (*M* age = 8.21y), 9M:6F. WCG: *N* = 15 (*M* age = 10.77y), 9M:6F. Groups matched for sex & duration of sleep problem. *Country:* U.K.	Sleep problems (NW, EMW, & settling)	RCT (int. vs WCG). *Follow up:* N/A.	*Agent:* Parent & child. *Int. Type:* Int. group received tailored behavioural sleep programmes based on a functional analysis. Incl.: Ext., graded Ext., stimulus control & Rx. *Dosage:* 3 mnths. *Clinical Support:* Both groups received 6 home visits. The int. group received additional ph calls (at least weekly), & could ph researcher at any time.	*Sleep Measures:* Activity monitors (worn by child & parent for usually 3 nights), parent report sleep diaries & parental questionnaire discussing child's sleep problems (all completed pre-int. (visit 1), during int. (visit 4) & post-int. (visit 6)). *Other Measures:* N/A.	*Parental Questionnaire: Child:* There was a sign. overall diff. between groups (F(1, 23) = 14.2, *P* = 0.001), an effect of time (F(2, 46) = 13.12, *P*<0.001), & a sign. interaction between the group & time (F(2,46) = 5.03, *P*<0.011). There was a diff. between pre-int. & visit 4 for the int. group & not for the WCG (t = 2.35, *P* = 0.027, 95% CL = 0.197 to 3.07), & between pre-int. & visit 6 (t = 3.50, *P* = 0.001, 95% CL = 0.758 to 2.94), but no further changes for either group between visits 4 & 6 (t = 3.14, *P* = 0.756, 95% CL = −1.18 to 1.61). *Activity Monitors: Child:* No diff. between groups, or diff. over time between groups. There was an effect of time on each variable (sleep period, activity score, movement index, & fragmentation index). Although no changes in children's objective sleep quality post-int., mothers in int. group reported improv. in the child's sleep problems, & ↑ TST.	*Treatment Fidelity:* N/A. *Social Validity:* N/A	Adequate
Willgerodt et al., 2014	*N* = 9, 8-11 y (*M* = 9.2), 5M:4F, children whose parent expressed TST concerns (4 had asthma). *Country:* U.S.	Variability in bedtime & TST.	Single-arm descriptive study. *Follow up:* N/A.	*Agent:* Parent & child. *Int. Type:* Ps completed 8 weeks of MBI which utilised the child's actigraphy data, goal setting tools & psychoeducation. *Dosage:* 6 weeks.. *Clinical Support*: 3 in-person visits (max 15mins each) & 2 mailed boosters (in between sessions). Reminder calls made mid-week to answer questions.	*Sleep Measures:* Sleep diaries (completed by parent & child together) & actigraphy (both for 7 days pre-int. (visit 1), for 14 days post-baseline (visit 2 & 3), & for 7 days 1 mnth later). *Other Measures:* ECBI (parent report) & researcher questions assessing the int. (parent & chid report), both at pre-int (visit 2) & 7 days later (visit 3).	*Actigraphy:* The 6 fully participating Ps (2 had asthma), showed ↓ in late bedtimes & bedtime variability. TST ↑ by 30min from pre-int to 2mnths later on >50% of the nights for 2 Ps, >42% of the nights for 2 Ps, & >25% of the nights for the other 2 Ps. *ECBI:* Av. parent-report child problem behavior scores ↓. 5/6 parents reported ↓ problem behavior freq. Parent report of how problematic the child's behaviour was ↓ or remained stable.	*Treatment Fidelity:* N/A. *Social Validity:* Ps with ADHD & learning disability reported to be unable to fully participate. Parents reported the clinical support of the int. acceptable & agreed that the int. was not burdensome.	Weak
Zhou et al., 2017	*N* = 10, 15-40y (*M* = 28.1y), 4M: 6F, cancer survivors experiencing insomnia & not in active cancer treatment,	Insomnia severity, daytime sleepiness, fatigue & QoL.	Pre-post case report. *Follow up:* 2 mnths post-int.	*Agent:* Adolescents & adults. *Int. Type:* 3x 60min individual behavioural int. sessions (10-14 days between sessions 1 & 2, & 10-20 days between sessions 2 & 3). Focus on behavioural components of CBT-I (sleep restriction &	*Sleep Measures:* Daily sleep diaries (from session 2). PSQI & ISI (both pre- & post-int. & f/up).	Stat. sign. improv. across all measures. *ISI:* *M* score ↓ to 6.3 (Cohens *d* = 1.0, *p* < .05) at post-int., & further ↓ to 4.2 (Cohens *d* = 1.3, *p* < .05) at f/up. *PSQI:* *M* score ↓ to 5.9 (Cohens *d* = 1.7, *p* < .05) at post-int. & to 3.4 (Cohens *d* = 2.3, *p* < .05) at f/up. *Sleep Diaries:* Improv. in SOL, NW's, & EMW.	*Treatment Fidelity:* N/A. *Social Validity:* Post-int. feedback: All Ps reported they would recommend the int. to other	Weak

					Other Measures: SF-12 (pre- & post-int. & f/up), & post-int. feedback questionnaire.	SF-12: ↑ in QoL scores (Cohens *d* = .3 to .7), but not stat. sign.	cancer survivors, that they did not regret participating, & that the int. helped them better understand their insomnia.	
	Survivors were Av. 12.5y post-Dx. & 10.9y post-treatment. *Country:* U.S.			stimulus control) & psychoeducation for sleep-related cognitions & sleep hygiene. *Dosage:* 20-34 days. *Clinical Support:* 6 Ps received all sessions in-person, 4 attended the 1st session in person & 2 via videoconference.				
Zhou & Recklitis, 2020	*N* = 22, 14-25y (*M* = 20.4y), 12M:10F, 77.3% non-Hispanic white ethnicity, cancer survivors (Av. 9.7y post-Dx.), with insomnia. *Country*: U.S.	Insomnia severity, daytime sleepiness, fatigue & QoL.	Single-arm efficacy trial (pre-post-test). *Follow up:* 8 weeks post-int (week 16).	*Agent:* Adolescents & adults. *Int. Type:* Ps accessed 6 sessions of an automated online program; Sleep Healthy Using The Internet (SHUTi). SHUTi utilised CBT-I strategies tailored for cancer survivors, & incl.: sleep restriction, stimulus control, cognitive therapy, sleep hygiene, & relapse prevention strategies, + sleep psychoeducation relating to cancer & suggestions for how to engage the P's family/friends in supporting the int. *Dosage:* Approx 6-8 weeks. *Clinical Support*: No research staff engagement once int. begun (SHUTi incl. interactive features & automated reminders).	*Sleep Measures* Daily sleep diaries (from session 2). ISI, PDSS, & PedsQL multidimensional fatigue scale (all at pre-int., post-int. (8 weeks), & f/up (16 weeks). *Other Measures:* PedsQL (pre-int, post-int. (8 weeks), & f/up (16 weeks).	*ISI:* Post-int., 50.0% of Ps reported ↓ by 6+ points, & at f/up this ↑ to 71.4% of Ps. Av. scores of Ps who completed 2+ sessions (n = 15) ↓ by 5.1 points between pre- to post-int., & ↓ by 10.0 points from pre-int. to f/up. Av. change from pre-to post-int. for those who completed 2+ sessions was sign. < than those who did not. The Av. scores of Ps who completed 3+ sessions (n = 12) ↓ from pre-int. to post-int. by 7.5 points, & by 10.9 points from pre-int. to f/up. *Sleep diaries:* Av. SE during 1st week = 80.3% (clinically indicative of insomnia). ↑ to Av. SE of 90.9% in session 6 (final session). *PDSS & PedsQL multidimensional fatigue scale:* Stat. sign. ↑ from pre- to post-int. (both Cohens *d* = 0.7, *P* < .05), maintained at f/up (PDSS: Cohens *d* = 1.5, *P* < .05, PedsQL multidimensional fatigue scale: Cohens *d* = 1.2, *P* < .05). *PedsQL:* Stat. sign. ↑ from pre-int to post-int. (Cohens *d* = 0.4, *P* < .05), maintained at f/up (Cohens *d* = 0.5, *P* < .05).	*Treatment Fidelity:* Ps completed an Av. of 3.2 sessions (3 Ps completed 0, & 6 completed all 6). *Social Validity:* N/A.	Weak
Zupanec et al., 2017	*N* = 20, 4-10y, 45% "white" ethnicity, all receiving maintenance chemotherapy for Acute Lymphoblastic Leukemia. Int. group: *N* = 11 (*M* age 6.3y), 10M:1F. CG: *N* = 9 (*M* age 6.2y), 8M:1F. *Country:* Canada.	Sleep disturbance & fatigue.	Pilot RCT (int. vs CG). *Follow up:* N/A.	*Agent:* Parent & child. *Int. Type:* Int. administered during groups 1st clinic visits of a new maintenance therapy cycle. Int. group received a 1-to-1 60min education session (covering sleep & fatigue in children with cancer), + strategies to ↑ sleep. incl. 2 storybooks to teach deep breathing & progressive muscle relaxation (Ps also given a brochure containing all info, a bedtime pass, & bedtime routine checklist). Ps were asked to implement the sleep hygiene & relaxation strategies over the next 4 weeks. The CG were not required to do anything beyond the usual clinic activities. *Dosage:* 4 weeks. *Clinical Support*: Within 1 week post-baseline, a ph call was made to review session & answer questions.	*Sleep Measures:* Actigraphy & sleep diaries (completed for 5 nights at pre- & post-int.). CCFS-C (Child report if child 7+ y), CCFS-P (Parent report if child <7 y), FISH, CSHQ. All completed pre- & post-int. *Other Measures:* Questionnaire rating the int. (parent report).	*Actigraphy:* Int. groups Av. night-time sleep ↑ by 35mins compared with the CG; but this diff. did not reach stat. sign. (*P* = .30). Int. group WASO ↓ by 44mins compared with the CG; this diff. almost reached stat. sign. (*P* = .08). Change from pre- to post-int. was similar across groups on day- time sleep duration, longest stretch of daytime & night-time sleep, & number of NW's. For > than 2/3's of Ps, parent-report bed & wake times did not allow enough time for children to achieve recommended amounts of sleep. *CCFS-P FISH & CSHQ:* No diff. between groups for pre- to post-int. scores (few Ps old enough to complete the CCFS-C, so data was not summarized). FISH pre- & post-int. scores were high (*M* score >46 in both groups), indicating that Ps were practicing good sleep habits prior to int.	*Treatment Fidelity:* 3 int. sessions were audiotaped so they could be reviewed for fidelity & consistency. Nurses completed an int. fidelity checklist during ph call at 1 week post-baseline to check components were being delivered. During weeks 1-4, sleep tips were used 5+ times per week by most of Ps (n = 7/8). *Social Validity:* All 8 Ps who completed int. evaluations rated the int. as useful & informative.	Adequate

Note. < = less than; > = greater than; ↑ = increase; ↓ = decrease; ADHD = Attention-Deficit/Hyperactivity Disorder; approx = approximately; ASHS = Adolescent Sleep Hygiene Scale; ASWS = Adolescent Sleep Wake Scale; ASWS-R = Adolescent Sleep Wake Scale-Revised; Av. = average; Ax. = assessment; BMA = Bone Marrow Aspirations; CALI-21 = Child Activity Limitations Interview-21; CBT= Cognitive Behavioral Therapy; CBT-I = Cognitive Behavioral Therapy for Insomnia; CSHQ: Child Sleep Habits Questionnaire; CCFS-C = Childhood Cancer Fatigue Scale-Child; CCFS-P = Childhood Cancer Fatigue Scale-Parent; CG = Control Group; CI = Confidence Interval; CRSP = Children's Report of Sleep Patterns; dev. = developed; diff. = difference; Dx. = diagnosis; ECBI = Eyberg Child Behavior Inventory; EMW = Early Morning Waking's; ES = Effect Size; Ext. = extinction; F = female; *F* = F statistic; FISH: Family Inventory of Sleep Habits; freq. = frequency; f/up: follow-up; GHQ = General Health Questionnaire; hr = hour; HRQOL= Health Related Quality Of Life; improv. = improved/improvements; incl. = including; int. = intervention; ISI = Insomnia Severity Index; M = male; *M* = mean; med = medium; min = minute; *N* = sample size; NW = Night Waking; N/A = Not Assessed; *P* = p value; PedsQL: Pediatric Quality of Life inventory; PDSS = Pediatric Daytime Sleepiness Scale; ph = phone; PROMIS = Patient Reported Outcomes Measurement Information System; Ps = participants; PSAS = Pre-sleep Arousal Scale; PSKI = Parents' Sleep Knowledge Inventory; PSQI = Pittsburgh Sleep Quality Index; psyc = psychological; QoL = Quality of Life; RCT= Randomised Control Trial; Rx = reinforcement; SD = Standard Deviation; SDI = Sleep Disturbance Index; SE = Sleep Efficiency; SF-12 = Short Form-12; SHI = Sleep Hygiene Index; sign. = significant; SOD = Sleep Onset Delay; SOL= Sleep Onset Latency; spec. = specified; stat. = statistically; TEI-SF = Treatment Evaluation Inventory, Short Form; TST = Total Sleep Time; vs = versus; WASO= Wake After Sleep Onset; y = years

Evaluation of Study Quality

The methodological quality of the 14 included articles was assessed by the thesis author using the evaluative method by Reichow et al. (2008; see Tables 3 and 4). The Reichow et al. (2008) criteria provides a standardised method for rating the empirical evidence of interventions for both group and Single-Case Research Designs (SCRD) and has been widely used in research (McLay et al., 2019; Hunter et al., 2020; McLay et al., 2020; Oudshoorn et al., 2020; Sun et al., 2020; Pervin et al., 2022). The methodological *rigor* of individual studies was rated according to primary and secondary indicators, as described below. This provided the basis for determining the research quality *strength* (i.e., strong, adequate or weak) of each study. The number of studies classified as meeting criteria for strong or adequate rigor, in conjunction with the treatment outcomes reported in each study (i.e., whether sleep problems reduced), was used to determine whether the body of research met criteria to be considered an established or promising evidence-based practice (Reichow et al., 2008).

Research Rigor. Depending on whether the study had a group design or SRCD, one of two different rubrics (guidelines) were used to assess the rigor of each study in relation to primary and secondary quality indicators. Primary quality indicators were deemed critical to the validity of a study and were defined according to a trichotomous scale (high quality, acceptable quality or unacceptable quality; Reichow et al., 2008). Primary quality indicators common to both group design and SCRD included evaluating: (a) whether sufficient detail was reported on participant characteristics (including participant age, gender, and diagnosis); (b) independent variables (whether there was sufficient information to enable replication); and (c) whether dependent variables were operationalized, replicable, relevant, and appropriate. Primary indicators specific to group designs included the study reporting: (a) comparison conditions; (b) alignment between research questions and data analysis; and (c)

Table 3

Quality Indicators for Group Studies

Study	**Ps**	**IV**	**Comp**	**DV**	**RQ**	**Stats test**	Random	IOA	Blind	Fidelity	Attrition	Main	ES	SV	Rating
Law et al., 2018	√√	√√	X	√√	√√	√	No	No	No	Yes	Yes	Yes	Yes	Yes	Weak
Meltzer & Booster, 2016	√	√√	√	√√	√√	√	No	No	No	No	No	No	No	Yes	Weak
Palermo et al., 2016, 2017	√√	√√	X	√√	√√	√	No	No	Yes	Yes	Yes	Yes	Yes	Yes	Weak
Sonney et al., 2020	√√	√√	X	√√	√√	√	No	No	No	No	Yes	Yes	Yes	Yes	Weak
Tsai et al., 2020	√√	√√	√√	√√	√√	√√	Yes	No	Yes	No	Yes	No	No	Yes	Strong
Tumakaka et al., 2019	X	X	√	√	√√	√	No	No	No	No	Yes	No	No	Yes	Weak
Wiggs & Stores, 1998	√	√√	√√	√√	√√	√	No	No	No	No	Yes	No	No	Yes	Adequate
Zhou et al., 2017	√	√	X	√√	√√	√	No	No	No	No	Yes	Yes	No	Yes	Weak
Zhou & Recklitis, 2020	√	√	X	√√	√√	√	No	No	No	No	No	Yes	Yes	Yes	Weak
Zupanec et al., 2017	√√	√√	√√	√√	√√	√	Yes	No	No	No	Yes	No	No	Yes	Adequate

Note. Primary indicators in bold; √√ = high quality; √ = acceptable quality; X = unacceptable quality; Ps = participant characteristics; IV = independent variable, comp = comparison condition; DV = dependent variable; RQ = link between research question and data analysis; stats test = use of statistical tests; random = random assignment; IOA = interobserver agreement; blind = blind raters; main = generalization and/or maintenance; ES = effect size; SV = social validity

Table 4

Quality Indicators for Single Case Studies

Study	**Ps**	**IV**	**DV**	**Base**	**Visual**	**Exp Con**	IOA	Fidelity	Blind	Main	SV	Rating
Bartlet & Beaumont, 1998	√√	X	X	X	X	X	No	No	No	Yes	Yes	Weak
Kellerman, 1979	√	√√	√√	X	√	X	No	No	No	No	Yes	Weak
Willgerodt et al., 2014	√√	√√	√√	X	X	X	No	No	No	No	Yes	Weak

Note. Primary indicators in bold; √√ = high quality; √ = acceptable quality; X = unacceptable quality; Ps = participant characteristics; IV = independent variable, DV = dependent variable; base = baseline condition; visual = visual analysis; exp con = experimental control; IOA = interobserver agreement; blind = blind raters; main = generalization and/or maintenance; SV = social validity

the use of statistical tests. Primary indicators distinct to SCRD included the study reporting: (a) a baseline condition; (b) visual analysis; and (c) experimental controls (Reichow et al., 2008).

Secondary quality indicators evaluate design quality features that are not necessarily essential for the validity of the study, however, still contribute to the overall design quality. These were defined according to a dichotomous scale (evidence or no evidence). Common secondary quality indicators to both group designs and SCRD included the study reporting: (a) interobserver agreement; (b) blind raters; (c) procedural fidelity; (d) generalisation and maintenance; and (e) social validity. Secondary indicators specific to group designs included the study reporting: (a) random assignment; (b) attrition; and (c) effect size (ES). There was also one secondary quality indicator specific to SCRD, which was the calculation of kappa (a statistic used to measure interobserver agreement [IOA]; Reichow et al., 2008).

Research Strength. The strength of each research study was classified according to the Reichow et al. (2008) criteria as follows; *strong* if the study demonstrated concrete evidence of high quality, *adequate* if the study demonstrated strong evidence in most but not all areas, or *weak* if the study had many missing elements, and/or fatal flaws. To assess the reliability of the ratings provided, inter-rater agreement was assessed for 20% of the studies included in this review (*n* = 4). These studies were randomly selected and independently evaluated by another member of the research team. The final agreement across ratings was 100%.

Effectiveness of the Evaluative Method. Two validation studies (Reichow et al., 2008; Cicchetti, 2011) have provided support for the reliability and validity of the evaluative method in assessing evidence-based practices in those with Autism. A review by Wendt and Miller (2012) also compared the compliance of seven quality appraisal tools designed to identify evidence-based practice with regard to current standards of conducting SCRD

research. Within their review, the evaluative method of Reichow et al. (2008) was reported as "very rigorous", as the use of primary and secondary quality indicators enabled clear identification of study strengths and weaknesses, consequently allowing users to easily distinguish between what constitutes strong, adequate, or weak research (Wendt & Miller, 2012). Further, the framework was rated as being of high quality due to its congruence with agreed standards for quality in SCRD. Additionally, of all the SCRD quality appraisal tools evaluated, Wendt and Miller (2012) reported that the evaluative method may be the most appropriate appraisal tool for use in comprehensive systematic reviews which aim to inform both clinical/educational practice and policy.

Results

Participants and Target Behaviours

The 13 intervention studies originally included a total of 485 participants, however the characteristics of only 474 participants were presented as three studies (Sonney et al., 2020; Wiggs & Stores, 1998; Zhou et al., 2017) exclusively reported the characteristics of participants who completed or began interventions. Five studies were affected by participant attrition (Bartlet & Beaumont, 1998; Law et al., 2018; Palermo et al., 2016, 2017; Tsai et al., 2020; Zupanec et al., 2017), with a total of 42 participants being lost between intervention and post-intervention or follow up assessments across these studies. The mean age of participants was 8.55 years (range 11 months – 40 years), and 50.2% were male. Eleven of the 13 studies had an upper age limit of 18 years, while the remaining two studies included participants up to 25 years (Zhou & Recklitis, 2020) and 40 years (Zhou et al., 2017). Two studies did not report their age ranges (Meltzer & Booster, 2016; Wiggs & Stores, 1998), and one study (Tumakaka et al., 2019) did not report the mean age of participants, nor gender. Eight studies were based in the U.S (Kellerman, 1979; Law et al., 2018; Meltzer & Booster, 2016; Palermo et al., 2016, 2017; Sonney et al., 2020; Willgerodt et al., 2014; Zhou

et al., 2017; Zhou & Recklitis, 2020), two in the U.K. (Bartlet & Beaumont, 1998; Wiggs & Stores, 1998), one in Taiwan (Tsai et al., 2020), one in Indonesia (Tumakaka et al., 2019) and one in Canada (Zupanec et al., 2017).

Four studies included participants with asthma (Bartlet & Beaumont, 1998; Meltzer & Booster, 2016; Sonney et al., 2020; Willgerodt et al., 2014), three studies included participants with a diagnosis of epilepsy (Bartlet & Beaumont, 1998; Wiggs & Stores, 1998; Tsai et al., 2020), two studies included participants with eczema/atopic dermatitis (Bartlet & Beaumont, 1998; Meltzer & Booster, 2016), acute lymphocytic leukaemia (Kellerman, 1979; Zupanec et al., 2017), or previous diagnoses of cancer (Zhou et al., 2017; Zhou & Recklitis, 2020), and only single studies included participants with chronic upper-respiratory tract infections (Bartlet & Beaumont, 1998), chronic migraine (Law et al., 2018), coeliac disease (Bartlet & Beaumont, 1998), or type 1 diabetes mellitus (Tumakaka et al.,2019).

In one study (Palermo et al., 2016, 2017), participants had co-occurring diagnoses of psychological (e.g., ADHD, anxiety, depression) and/or physical conditions (e.g., asthma/eczema, chronic pain). In one study (Bartlet & Beaumont, 1998), participants either had neurodevelopmental disorders (e.g., autism, down syndrome, specific learning disability) or a chronic illness (e.g., eczema, asthma, epilepsy). In one study (Wiggs & Stores, 1998) participants were reported to have "at least one form of daytime challenging behaviour", this included children with neurodevelopmental disorders (e.g., autism, down syndrome) and/or epilepsy. Finally, in the study by Willgerodt et al. (2014) only four of the nine participants had a diagnosis of asthma, and three participants had a developmental disorder.

All participants across the 13 studies experienced at least one type of sleep problem, which included problems with bedtime settling, sleep onset delay, NW, Early Morning Waking (EMW) and daytime fatigue. The most common targeted sleep problems, were

overall sleep quality (measured by the Pittsburgh Sleep Quality Index [PSQI] or sleep

diaries), insomnia, and fatigue which were all each discussed by three studies.

In four studies, participants were simultaneously receiving sleep medications during

psychosocial interventions. Specifically, in the study by Bartlet and Beaumont (1998), 63%

of participants ($n = 39$) had received hypnotics to promote sleep, 38% ($n = 23$) had received

vallergan (trimeprazine), and 36% ($n = 22$) had received phenergan (promethazine). In the

study by Law et al. (2018), 19% ($n = 4$) were taking prescription sleep medications and

33.3% ($n = 7$) were taking over the counter sleep medications, including melatonin (42.8%).

In the study by Palermo et al. (2016, 2017), 12.5% ($n = 5$) were taking prescription sleep

medications and 27.5% ($n = 11$) were taking over the counter sleep medications. Finally, in

the study by Zhou and Recklitis (2020), 36.4% ($n = 8$) were taking a medication for sleep

(clonazepam, mirtazapine, hydroxyzine, melatonin, and diphenhydramine).

Study Design and Follow-up

Of the 13 studies, ten employed a group design and the remaining three were SCRD.

Of the group design studies, three (Tsai et al., 2020; Wiggs & Stores, 1998 Zupanec et al.,

2017) used a Randomised Controlled Trial (RCT; sample size ranged from 20 – 100

participants, $M = 50$). One used a non-randomised controlled study design (Meltzer &

Booster 2016; $n = 89$), one study used a pre-post quasi-experimental design with a non-

equivalent control group (Tumakaka et al., 2019, $n = 46$), and five (Law et al., 2018; Palermo

et al., 2016, 2017; Zhou et al., 2017; Sonney et al., 2020; Zhou & Recklitis, 2020) utilised a

pre/post design with no control group (sample size ranged from 10 – 40 participants, $M = 23$).

Of the SCRD, one study (Bartlet & Beaumont, 1998) used a nonconcurrent multiple baseline

across participants design ($n = 61$), one (Kellerman, 1979) was a case study ($n = 1$), and one

(Willgerodt et al., 2014) was a single-arm descriptive study ($n = 9$). Six studies (Bartlet &

Beaumont, 1998; Law et al., 2018; Palermo et al., 2016, 2017; Sonney et al., 2020; Zhou et

al., 2017; Zhou & Recklitis, 2020) gathered follow-up data from eight weeks to six months post-intervention, only one of which was a SCRD study (Bartlet & Beaumont, 1998) where there was a three to six month follow-up.

Intervention Characteristics

In nine studies intervention was directed toward the child/adolescent and the parent together through collaborative strategies (Kellerman, 1979; Bartlet & Beaumont, 1998; Law et al., 2018; Palermo et al., 2016, 2017; Sonney et al., 2020; Tumakaka et al., 2019; Wiggs & Stores, 1998; Willgerodt et al., 2014; Zupanec et al., 2017). Three of these studies (Kellerman, 1979; Bartlet & Beaumont, 1998; Wiggs & Stores, 1998) utilised individualised behavioural strategies to target specific sleep difficulties the child was experiencing. Within these studies the following techniques were used: cuing (using specific stimuli to set the conditions for sleep), graded change (breaking down the sequence of change into discrete steps), stimulus control, extinction, graduated extinction, and reinforcement strategies. One study (Willgerodt et al., 2014) used a developmentally tailored Motivation-Based Intervention (MBI), where children and parents reviewed the child's actigraphy data, set goals, discussed barriers to their goals and were provided with sleep education. Two studies (Zupanec et al., 2017; Tumakaka et al., 2019) solely used sleep hygiene education, with Zupanec et al. (2017) also including sleep hygiene education related to cancer. One study (Sonney et al., 2020) implemented a program developed by their research team; Sleep Intervention for Kids & Parents (SKIP). SKIP involved a researcher providing baseline actigraphy feedback to the parent and child, and then recommending online SKIP modules. These modules focused on behavioural strategies, elements of CBT-I and goal setting tasks related to bedtime routine, sleep quality and the bedtime environment.

Four studies (Palermo et al., 2016, 2017; Zhou et al., 2017; Law et al., 2018; Zhou & Recklitis, 2020) utilized CBT-I (i.e., sleep hygiene education, stimulus control, and sleep

restriction). Three of these studies (Palermo et al., 2016, 2017; Law et al., 2018; Zhou & Recklitis, 2020) also specified relating information to the specific CHC. Law et al. (2018) added components related to pain management (e.g., headache education, relaxation training, pleasant activity scheduling and positive thought tracking), parent operant training to reinforce the skills adolescents were taught, and three optional modules on anxiety management, fatigue management, and activity pacing. Palermo et al. (2016, 2017) also included optional treatment strategies (e.g., strategies to address bed- time resistance and co-sleeping [planned parental ignoring], relaxation strategies, strategies to address problematic sleep cognitions [cognitive restructuring], strategies to address fatigue, and social support for sleeping away from home). Both Law et al. (2018) and Palermo et al. (2016, 2017) utilized a collaborative format which included the adolescent's parents throughout sessions, whereas Zhou et al. (2017) and Zhou and Recklitis (2020) solely worked with the adolescent or adult directly. Zhou and Recklitis's (2020) developed their own automated online program; Sleep Healthy Using The Internet (SHUTi) which included CBT-I as well as specific sleep education relating to cancer and insomnia.

Two studies were directed toward the parent and focused on changing parental behaviours as a means of addressing sleep problems in their child (Meltzer & Booster, 2016; Tsai et al., 2020). Meltzer and Booster (2016) provided a group intervention which involved sleep education (e.g., education on developmental sleep needs, the impact of chronic illnesses) and strategies to increase sleep. Tsai et al. (2020) directed a program they had developed toward parents called Sleep, Health, and Research for Epilepsy (SHARE). SHARE included using the children's actigraphy data to provide parents with specific feedback, sleep education and strategies to promote sleep, as well as discussions regarding goals and potential barriers to achieving them.

Five studies (Meltzer & Booster, 2016; Palermo et al., 2016, 2017; Zupanec et al., 2017; Law et al., 2018; Tsai et al., 2020) provided additional resources (e.g., handouts, story books, bedtime pass, bedtime routine checklist), with two studies including manuals (Palermo et al., 2016, 2017; Law et al., 2018). Three studies provided homework for children/adolescents to complete (Palermo et al., 2016, 2017; Law et al., 2018; Sonney et al., 2020).

In terms of intervention delivery, seven studies implemented interventions in person at the clinic or research institute (Kellerman, 1979; Law et al., 2018; Meltzer & Booster, 2016; Palermo et al., 2016, 2017; Tsai et al., 2020; Willgerodt et al., 2014l Zupanec et al., 2017). Three studies implemented interventions in either the participants home or the paediatric outpatient department (Bartlet & Beaumont, 1998; Sonney et al., 2020; Zhou et al., 2017). Sonney et al. (2020) using a combination of clinic visits and online modules, and Zhou et al. (2017) giving participants the option of in person or videoconference delivery. One intervention was delivered in person at the family's home (Wiggs & Stores, 1998), one was delivered using a fully online program (Zhou & Recklitis, 2020), and one was solely delivered via video (Tumakaka et al., 2019).

Intervention dosage for treatment groups varied across studies and ranged from one to six sessions, with the exception of one study (Kellerman, 1979) where sessions ran twice weekly and gradually reduced to weekly, fortnightly and monthly over 246 days. The sessions within interventions also varied in duration from 10 minutes to two hours. Four studies (Kellerman, 1979; Bartlet & Beaumont, 1998; Wiggs & Stores, 1998; Willgerodt et al., 2014; Zupanec et al., 2017) also provided additional support/reminder calls and/or boosters. The boosters involved either an additional in person session following the final treatment session (Law et al., 2018), or mailed correspondence during the intervention between the in person sessions (Willgerodt et al., 2014). One study (Zhou & Recklitis, 2020)

did not provide any clinical support in addition to the online program, and in two studies (Bartlet & Beaumont, 1998; Tsai et al., 2020) the duration of treatment was unclear.

The duration of interventions also varied from one day (Meltzer & Booster, 2016; Tumakaka et al., 2019) to 12 months (Tsai et al., 2020). The study by Tsai et al. (2020) involved the longest intervention process where participants engaged in face to face sessions at baseline, three- and six-months, followed by a phone call at nine-months which involved further clinical support. Majority of the studies (*n* = 9) involved clinical support and/or programme activities which lasted approximately one to three months (Bartlet & Beaumont, 1998; Law et al., 2018; Palermo et al., 2016, 2017; Sonney et al., 2020; Wiggs & Stores, 1998; Willgerodt et al., 2014; Zhou et al., 2017; Zhou & Recklitis, 2020; Zupanec et al., 2017).

Sleep Measures

Sleep measures included psychometric questionnaires, sleep diaries, and actigraphy. Nine studies (Bartlet & Beaumont, 1998; Meltzer & Booster, 2016; Palermo et al., 2016, 2017; Zhou et al., 2017; Zupanec et al., 2017; Law et al., 2018; Tumakaka et al., 2019; Tsai et al., 2020; Zhou & Recklitis, 2020) used established sleep questionnaires at pre- and post-intervention, the most common being the Insomnia Severity Index (ISI; used by Law et al., 2018; Meltzer & Booster, 2016; Palermo et al., 2016, 2017; Zhou et al., 2017; Zhou & Recklitis, 2020), and the Pittsburgh Sleep Quality Index (PSQI; used by Tsai et al., 2020; Tumakaka et al., 2019; Zhou et al., 2017). Additionally, one study (Wiggs & Stores, 1998) used the Modified Simonds and Parraga Sleep Questionnaire (MSPSQ; Simonds & Parraga 1982) to inform the development of their own questionnaire.

In four of the nine studies (Law et al., 2018; Palermo et al., 2016, 2017; Zhou et al., 2017; Zhou & Recklitis, 2020) psychometrics were repeated at follow-up. Psychometric sleep measures included both parent (Bartlet & Beaumont, 1998; Meltzer & Booster, 2016; Tsai et

al., 2020; Tumakaka et al., 2019; Zupanec et al., 2017) and self-report (Law et al., 2018; Palermo et al., 2016, 2017; Zhou et al., 2017; Zhou & Recklitis, 2020; Zupanec et al., 2017).

Sleep diaries were used in 11 studies, and included parent-report (Bartlet & Beaumont, 1998; Kellerman, 1979; Tsai et al., 2020; Wiggs & Stores, 1998), and self-report (Law et al., 2018; Palermo et al., 2016, 2017; Zhou et al., 2017; Zhou & Recklitis, 2020). In three studies (Sonney et al., 2020; Willgerodt et al., 2014; Zupanec et al., 2017) the sleep diaries were completed collaboratively by the parent and child. Sleep diaries were typically used to measure bedtime range and variability, Sleep Onset Latency (SOL), Wake After Sleep Onset (WASO), NW, EMW, SE and TST, with one study also measuring night terrors (Kellerman, 1979). The duration and frequency of sleep diary recording varied across studies. In three studies (Kellerman, 1979; Zhou et al., 2017; Zhou & Recklitis, 2020), participants were asked to record for a majority or the full duration of the study, while in the remaining studies sleep diaries were recorded for five nights at pre- and post-intervention (Zupanec et al., 2017), or for seven or 10 days at three different time points (Law et al., 2018; Palermo et al., 2016, 2017; Sonney et al., 2020; Tsai et al., 2020; Wiggs & Stores, 1998; Willgerodt et al., 2014).

Of the 11 studies which used sleep diaries, six studies also used actigraphy (Palermo et al., 2016, 2017; Sonney et al., 2020; Tsai et al., 2020; Wiggs & Stores, 1998; Willgerodt et al., 2014; Zupanec et al., 2017) to measure sleep. This provided an objective measure of SE, TST, WASO, NW, movement and activity, and sleep fragmentation, which supplemented sleep diary findings. Participants wore actigraphy for three, five, seven or 10 days at either two (Zupanec et al., 2017), or three (Palermo et al., 2016, 2017; Sonney et al., 2020; Tsai et al., 2020; Wiggs & Stores, 1998; Willgerodt et al., 2014) time points.

Additional Outcome Measures

In addition to sleep, five studies (Willgerodt et al., 2014; Palermo et al., 2016, 2017; Zhou et al., 2017; Law et al., 2018; Zhou & Recklitis, 2020) assessed non-sleep related outcomes. These included measures of children's physical health (e.g., headache diary, the Child Activity Limitations Interview-21 [CALI-21]; Law et al., 2018, The Patient Reported Outcomes Measurement Information System [PROMIS]; Palermo et al., 2016, 2017), mental health (e.g., Paediatric Anxiety Short Form and Paediatric Depressive Symptoms Short Form; Palermo et al., 2016, 2017), quality of life (e.g., the Paediatric Quality of Life Inventory [PedsQL]; Palermo et al., 2016, 2017; Zhou & Recklitis, 2020), health-related quality of life (e.g., the Short Form-12 [SF-12]; Zhou et al., 2017), and daytime behaviour (e.g., the Eyberg Child Behavior Inventory [ECBI]; Willgerodt et al., 2014).

Treatment Fidelity

Treatment fidelity was reported to be assessed in five studies (Law et al., 2018; Palermo et al., 2016, 2017; Tsai et al., 2020; Zhou & Recklitis, 2020; Zupanec et al., 2017), however Tsai et al. (2020) did not report their fidelity results. Fidelity was typically assessed through regular researcher observations and/or reporting of participants' completion of intervention components. Of the four studies which reported treatment fidelity results, it was generally reported to be high in three of these studies (Palermo et al., 2016; Zupanec et al., 2017; Law et al., 2018). Specifically, Law et al. (2018) reported participants were highly compliant with homework completion (*M* homework completed= 9.50/10, SD = .74) and diary completion (average of 5/7 diary days reported at each assessment point), and Zupanec et al. (2017) reported that their sleep tips were used at least five times per week by majority of participants. One study (Zhou & Recklitis, 2020) did however report variability in completion of sessions, with three participants completing zero sessions, and six completing all six sessions.

Social Validity

Treatment acceptability was reported in eight studies (Bartlet & Beaumont, 1998; Law et al., 2018; Palermo et al., 2016, 2017; Sonney et al., 2020; Willgerodt et al., 2014; Zhou et al., 2017; Zupanec et al., 2017; Tsai et al., 2020), and was assessed using post-intervention feedback questionnaires, surveys and semi-structured interviews, and the Treatment Evaluation Inventory, Short Form (TEI-SF). Survey and interview outcomes indicated high levels of satisfaction with treatment procedures across the majority of participants. For example, interventions were rated as acceptable (Willgerodt et al., 2014; Zhou et al., 2017; Sonney et al., 2020; Tsai et al., 2020), useful (Bartlet & Beaumont, 1998; Zhou et al., 2017; Zupanec et al., 2017) and informative (Zhou et al., 2017; Zupanec et al., 2017).

Results of the TEI-SF, which were reported by two studies (Palermo et al., 2016; Law et al., 2018), showed an overall moderate level of treatment satisfaction, as rated by parents and adolescents. Law et al. (2018) and Palermo et al. (2016) reported an average parent score of 41 and 39 out of a possible 45, respectively, and an average adolescent score of 39 and 37 out of 45, respectively. Palermo et al. (2016) also reported specific results of TEI-SF items, which showed that parents and adolescents were receptive to learning the new skills and had been using them at home, found the intervention easy to understand, would be highly likely to recommend the treatment to others, and that the intervention was likely to result in long-lasting changes.

Sleep Intervention Outcomes

Overall, 12 of the 13 studies reported an improvement in at least one aspect of children's sleep, following intervention. Specifically, reductions in settling difficulties (Bartlet & Beaumont, 1998), SOL (Law et al., 2018; Palermo et al., 2017; Zhou et al., 2017), WASO (Law et al., 2018; Palermo et al., 2017; Sonney et al., 2020), NW (Bartlet &

Beaumont, 1998; Zhou et al., 2017), night terrors (Kellerman, 1979), EMW (Zhou et al., 2017) and daytime fatigue (Zhou & Recklitis, 2020) were reported. Three studies (Law et al., 2018; Palermo et al., 2017; Sonney et al., 2020) reported improvements in sleep hygiene, including regularity of bedtime (Sonney et al., 2020), and pre-sleep arousal and sleep/wake scheduling (Palermo et al., 2017). Improvements in overall SE (Law et al., 2018; Palermo et al., 2017; Sonney et al., 2020; Tsai et al., 2020; Zhou & Recklitis, 2020), TST (Sonney et al., 2020; Tsai et al., 2020; Wiggs & Stores, 1998; Willgerodt et al., 2014), sleep quality (Law et al., 2018; Tumakaka et al., 2019; Zhou et al., 2017) and a general reduction of insomnia symptoms (Wiggs & Stores, 1998; Law et al., 2018; Palermo et al., 2017; Zhou et al., 2017; Zhou & Recklitis, 2020) were also observed. Improvements in regularity of bedtime, WASO, SE and TST were also reflected in actigraphy data (Sonney et al., 2020; Tsai et al., 2020; Willgerodt et al., 2014; Zupanec et al., 2017). In the five studies that compared an intervention group with a control group (Wiggs & Stores, 1998; Meltzer & Booster, 2016; Tumakaka et al., 2019; Zupanec et al., 2017; Tsai et al., 2020), three studies (Wiggs & Stores, 1998; Tumakaka et al., 2019; Tsai et al., 2020) found significant between-group differences, one study (Zupanec et al., 2017) reported greater improvements for the intervention group, however this was not statistically significant, and the other study (Meltzer & Booster) reported no differences between the intervention and control group post-intervention.

It is important to note that there were many studies that reported varying levels of intervention effects across participants and sleep outcomes. For example, Palermo et al. (2017) reported TST was unchanged across assessments, Sonney et al. (2020) reported improvements in sleep variables except for bedtime variability, and Tsai et al. (2020) found no difference in daily sleep duration (as measured by actigraphy), nor the Children's Sleep Habits Questionnaire (CSHQ), or Pittsburgh Sleep Quality Index (PSQI) between the

intervention and control groups. Similarly, Zupanec et al. (2017) found no difference in scores on the Childhood Cancer Fatigue Scale-Parent (CCFS-P), Family Inventory of Sleep Habits (FISH) and CSHQ between the intervention and control group from pre- to post-intervention. Zupanec et al. (2017) also found no difference in improvement post-intervention between groups for day-time sleep duration, longest stretch of daytime and night-time sleep, and frequency of NW (Zupanec et al., 2017). Interestingly, in two studies (Wiggs & Stores, 1998; Palermo et al., 2017) there were differences in intervention outcomes depending upon the sleep measures used. For example Wiggs and Stores (1998) found that mothers in the intervention group reported reductions in their child's sleep problems and increased TST, however, actigraphy results showed no change in children's sleep quality. Additionally, Palermo et al. (2017) reported that unlike improvements found in WASO and SE (as reported by sleep diaries), few objective changes were detected with actigraphy. Finally, one study (Meltzer & Booster, 2016), found that post-intervention there were no significant differences in improvements between the intervention and the control group. For example, both groups reported improvements in regard to activities in the hour before bed, sleep location, the use of electronics at sleep onset and the Insomnia Scale regardless of their intervention participation; therefore, improvements could not be attributed to intervention.

Additional Intervention Outcomes

Two studies (Zhou et al., 2017; Zhou & Recklitis, 2020) reported improvements in QoL from pre- to post-intervention, with these improvements being significant for Zhou and Recklitis (2020). Palermo et al. (2017) also reported that while there were no changes in total QoL or physical & psychosocial QoL from pre- to post-intervention, there were significant reductions in symptoms of anxiety and depression on the Paediatric Anxiety Short Form and Paediatric Depressive Symptoms Short Form for adolescents. Willgerodt et al. (2014) reported that the average child problem behaviour scores decreased following intervention,

including the frequency and severity of problem behaviours decreasing or remaining stable, as reported by parents. Finally, Law et al. (2018) reported a significant reduction in headache frequency for adolescents, which was correlated with a decrease in insomnia symptoms. However, headache pain intensity and the activity limitations remained unchanged from pre- to post-intervention.

Maintenance of Treatment Effects

For the six studies (Law et al., 2018; Palermo et al., 2017; Sonney et al., 2020; Tsai et al., 2020; Zhou et al., 2017; Zhou & Recklitis, 2020) who included follow up assessments, the majority of treatment gains were maintained. Sleep treatment effects were found to be maintained for SOL (Law et al., 2018; Palermo et al., 2017), WASO (Law et al., 2018; Palermo et al., 2017; Sonney et al., 2020), and daytime fatigue (Zhou & Recklitis, 2020) at follow up. Sleep hygiene improvements were also maintained at follow up (Law et al., 2018; Palermo et al., 2017; Sonney et al., 2020), including regularity of bedtime (Sonney et al., 2020), and pre-sleep arousal and sleep/wake scheduling (Palermo et al., 2017). Improvements in overall SE (Law et al., 2018; Palermo et al., 2017; Sonney et al., 2020; Tsai et al., 2020), TST (Tsai et al., 2020), sleep quality (Law et al., 2018; Zhou et al., 2017), and a general reduction in insomnia symptoms (Law et al., 2018; Palermo et al., 2017; Zhou et al., 2017; Zhou & Recklitis, 2020) were also maintained. One exception was that improvements in TST were not sustained in the study by Sonney et al. (2020), with scores returning to pre-intervention levels. Treatment effects for other (non-sleep) outcomes were also found to be maintained for anxiety and depressive symptoms (Palermo et al., 2017), headache frequency (Law et al., 2018) and QoL (Zhou & Recklitis, 2020).

In two studies (Palermo et al., 2017, 2017; Law et al., 2018), further improvements were observed between intervention and follow-up. Law et al. (2018) reported improvements in TST and in the limitation of daytime activities at follow-up despite these variables

remaining stable from pre- to post-intervention. Additionally, Palermo et al. (2016; 2017) reported a significant difference in total QoL scores from pre-intervention to follow-up, but not from pre- to post-intervention..

Study Quality

Only one study (Tsai et al., 2020) met criteria to be classified as 'strong'. Two studies (Wiggs & Stores, 1998; Zupanec et al., 2017) were classified as having 'adequate' strength, and the remaining ten studies were classified as 'weak'. This included the one study which did not report improvements following intervention (Meltzer & Booster, 2016).

With regard to the primary indicators common to both group and SCRD studies, the majority of studies received acceptable or high quality ratings for participant characteristics, and independent variables (e.g., participants and treatment described with sufficient detail) except for two studies (Bartlet & Beaumont, 1998; Tumakaka et al., 2019), where participant characteristics were not described (Tumakaka et al., 2019), and/or the content of interventions was not provided in enough detail (Bartlet & Beaumont, 1998; Tumakaka et al., 2019). With regard to dependent variables, 12 studies received high quality ratings, with only one study (Bartlet & Beaumont, 1998) not reporting when all measures were completed. Finally, all 13 studies met criteria for social validity, however no studies reported IOA, only two studies (Palermo et al., 2016, 2017; Tsai et al., 2020) reported blinding, and only two studies (Law et al., 2018; Palermo et al., 2016; 2017) met criteria for fidelity.

For the ten group design studies, four studies (Law et al., 2018; Palermo et al., 2016, 2017; Sonney et al., 2020; Zhou et al., 2017; Zupanec et al., 2017) scored poorly due to not having a comparison group. With regard to statistical power, most studies did not mention statistical power at all, and only one study acknowledged their study was under-powered (Zupanec et al., 2017). Additionally, five studies (Wiggs & Stores, 1998; Meltzer & Booster, 2016; Zupanec et al., 2017 Tumakaka et al., 2019; Tsai et al., 2020) did not report effect size,

and two studies (Meltzer & Booster, 2016; Zhou & Recklitis, 2020) did not discuss attrition which made it difficult to report on these indicators and resulted in these studies scoring low on these respective quality indicators. Additionally, only two studies (Zupanec et al., 2017; Tsai et al., 2020) reported random assignment.

For the three SCRD studies, one study (Kellerman, 1979) met criteria for visual analysis, however no studies met criteria for baseline or experimental control. This was due to many factors including a lack of adequate baseline and/or visual analysis reporting and/or no replication.

Overall, in regard to research rigor and intervention outcomes the empirical evidence of the 13 studies, met criteria for the interventions to be classified as promising Evidence-Based Practice (EBP; Reichow et al., 2008).

Discussion

The present review sought to identify and evaluate research examining the effectiveness of psychosocial sleep interventions for children with CHC. Fourteen articles representing 13 studies met inclusion criteria. These studies included participants with a variety of CHC including asthma (Bartlet & Beaumont, 1998; Meltzer & Booster, 2016; Sonney et al., 2020; Willgerodt et al., 2014), acute lymphoblastic leukemia (Kellerman, 1979; Zupanec et al., 2017), and epilepsy (Bartlet & Beaumont, 1998; Tsai et al., 2020; Wiggs & Stores, 1998). Although the 14 studies were published between 1979 and 2020, a majority were published in the last ten years. This suggests that sleep problems in children with CHC is a growing area of research and instills hope that this may become an area of increased focus in the coming years.

Overall, twelve studies reported at least some positive effects of the psychosocial sleep interventions across participants. This included reductions in SOL, WASO, NWs, EMW and improvements in sleep hygiene and overall sleep quality. These improvements

were evident in subjective report (e.g., parental, child, adolescent or adult report of sleep diaries or psychometrics), though objective measures suggested some variability in outcomes (e.g., actigraphy). Improvements in QoL, daytime problem behaviours, symptoms of anxiety and depression, and headache frequency (for children with chronic migraine) were also reported following intervention. The majority of these improvements in sleep and collateral outcomes, were found to be maintained at short and/or long-term follow-up. In all seven studies reporting social validity outcomes, overall satisfaction with treatment procedures was reported to be moderate to high, and of the four studies which reported treatment fidelity, three reported this was generally high. The the findings of this review may therefore provide preliminary evidence of the effectiveness, acceptability and feasibility of psychosocial interventions for sleep problems in children with CHC.

Although some studies reported mixed findings (i.e., improvements were not seen across all sleep outcomes) or variability across measures (e.g., improvements reported by sleep diaries but not actigraphy), there was only one study (Meltzer & Booster, 2016) where no improvements in sleep outcomes were seen. In the study by Meltzer and Booster (2016) no significant between group differences were found between the intervention and control group. Interestingly, this study delivered their sleep health education via a one-time group intervention to the parents of children with atopic dermatitis and asthma. Meltzer and Booster (2016) suggest that the similar improvements seen between their intervention and control group, regardless of intervention participation, may have been due to the fact that children in both groups were partaking in a medical treatment program (for their CHC) which concluded prior to post-treatment assessment. Therefore, it was possible that as the children's health improved in relation to the medical treatment program, their sleep also improved regardless of their participation in the sleep intervention (Meltzer & Booster, 2016).

Additionally, although not discussed by Meltzer and Booster (2016), it is also possible that sleep education on its own, especially in a one-off format, was not sufficient to overcome sleep problems. This notion is supported by previous research where many sleep education sessions have been required for sleep to improve (Blunden, 2007a; Blunden, 2007b; Kira et al., 2014; Rigney et al., 2015; Gruber et al., 2016; Illingworth et al., 2020). Additionally, in a review by Blunden et al. (2012) of sleep education programs for typically developing children, despite most programs resulting in increases in sleep knowledge, increases in sleep knowledge did not necessarily also result in changes in sleep behaviours (e.g., changes not seen in sleep duration or improved sleep hygiene). Blunden et al. (2012) proposed that this lack of change may have been due to a lack of motivation and readiness to change, as well as the delivery, content and time allocation of the program.

These limitations of short sleep education programs were also discussed in a systematic review by Chung et al. (2017) who investigated school-based sleep education programs for adolescents (aged 10 – 19). Chung et al. (2017) highlighted that school-based sleep education programs provided short-term benefits, however improvements were often not maintained over time. Chung et al. (2017) concluded that the content, duration and parental involvement of the programs may have impacted their relative effectiveness. Thus, it is important to consider how short sleep education interventions, such as that by Meltzer and Booster (2016) may not be sufficient to provide long-lasting sleep improvements.

Only one study in this review met criteria to be considered 'strong'. The remaining studies were classified as 'adequate' (2/13 studies) or 'weak' (10/13 studies). Common limitations to these studies which impacted their quality included a lack of visual analysis, a lack of reliability (IOA) and treatment fidelity checks, limited blinded data collection and an insufficient degree of experimental control due to only five studies including control groups. However, it is important to note that many of the studies were exploratory in nature as they

were investigating a new area of research. In spite of the methodological limitations, the use of psychosocial sleep interventions with children with CHC still emerged as promising EBP according to the Reichow et al. (2008) criteria.

In general, interventions were provided to either parents alone, the adolescent/adult alone, or to parents and their children/adolescents in combination. Sleep education was provided regarding the factors that contribute to and maintain sleep problems, as well as behavioural strategies to respond to sleep-competing behaviours. A wide range of interventions were used across studies. Many of these included common sleep intervention strategies used within typically developing children, such as behavioural sleep strategies and CBT-I. Various reviews have reported behavioural sleep interventions to be effective in improving sleep within typically developing children and children with developmental disabilities (Mindell, 1999; Owens et al., 1999; Wiggs & France, 2000; Mindell et al., 2006; Meltzer & Mindell, 2014; Rigney et al., 2018; Carnett et al., 2020; Pattison et al., 2020). Two previous reviews have also reported that CBT-I may be effective in typically developing children (Blake et al., 2017; Ma et al., 2018). Thus the current review provides preliminary evidence to suggest that psychosocial sleep interventions which are effective in typically developing children and children with developmental disabilities, are also effective for children who face a different set of physiological and psychological challenges.

Importantly, it is possible that the specific use of CBT-I strategies within five of the included studies was effective as they provided a means for the intervention to be tailored to the specific contributing factors of the CHC (e.g., CBT including pain management). Because it is known that children with CHC are at a higher risk of psychological difficulties, and that there is a strong association between sleep and psychological functioning, these strategies may have therefore enabled interventions to target various psychological factors that contribute to sleep difficulties in children with CHC.

Incorporating sleep education related to the specific CHC may have also been an important component of the interventions. Sleep education may also help to emphasise the importance of sleep for the child's physical health, which may result in increased motivation. Including a focus on the child's CHC also allowed these studies to take a holistic approach which didn't distinguish physical health from sleep health. Previous research within the care of children with various CHC has emphasised the importance of a multidisciplinary approach which considers all aspects of health (Warner et al., 1989; Shute, 1997; Chan et al., 2001; Carlson et al., 2008; Zolotarjova et al., 2018; Leach et al., 2021; Cantrell et al., 2022). Thus it is clear that an approach which is tailored to the specific CHC and emphasises the overall health of the child, while considering how other factors (e.g., medical management of the condition) impact sleep, is important for planning effective sleep interventions in children with CHC.

In the majority of included studies, intervention agents included both the child/adolescent and the parent(s). Utilising collaborative approaches is beneficial within paediatric sleep interventions as it enables the child to maintain a sense of control and autonomy, while also ensuring that learnt strategies are implemented in the home environment through actions from the parents that further reinforce appropriate sleep behaviours. Including parents may also help parents to learn to modify their responses to their child's behaviours in order to avoid reinforcing sleep interfering behaviours. Importantly, parents may often change their expectations around sleep, and consequently their responses to sleep interfering behaviours when their child has a CHC. Therefore providing education on the importance of sleep may help to ensure parents understand the importance of sleep to their child's overall health.

For older children taking a collaborative approach may also allow children to continue their journey towards becoming in control of managing their illness, while

maintaining parental support to ensure intervention strategies are reinforced. This is important as previous research has emphasised the important transition which occurs toward adolescence, where children begin to have increased responsibility for managing their symptoms and overall health (Christian et al., 1999; Sawyer & Aroni, 2005; Reed-Knight, 2014; Lerch & Thrane, 2019). For example, as a child with diabetes ages they may be given more responsibility for self-administering their insulin injections (Rankin et al., 2018). Sleep interventions that include older children as agents, giving them an active role within their own treatment, may therefore help to establish lifelong healthy sleep behaviours as part of their independent health care.

Another important finding of the current review which warrants further investigation is the differences in outcomes between parent- or self-report and actigraphy. Palermo et al. (2017) found that the moderate to large changes captured by parent- and self-report data were not consistently reflected in actigraphy data. The authors suggested that this could reflect several issues related to using actigraphy in the context of a brief intervention. A review on the use of actigraphy for assessment in pediatric sleep by Meltzer et al. (2012) found actigraphy was consistently reported to have poor specificity in detecting WASO across devices and age groups. Additionally, as parent- and self-report (e.g., sleep diaries) reflect the individual's perception of sleep (e.g., whether they or the child slept poorly), CBT-I interventions may be more likely associated with parent- and self-reported improvements, rather than objective improvements (Palermo et al., 2017). This is because CBT-I aims to correct dysfunctional beliefs or misperceptions about sleep, which may change how an individual perceives and therefore reports their sleep to be, compared to actigraphy which may not capture the improvements from changes in sleep cognitions as quickly (Palermo et al., 2017).

Another explanation for why there may have been differences between subjective and objective report was given by Wiggs and Stores (1998) who proposed that treatment may have reduced children's signaling of their awake state to their parents, rather than affecting the children's sleep quality or quantity per se. This may help to explain why parent-reported improvement were not reflected in actigraphy in some cases. Thus, it is possible that children's sleep did not actually improve, however parents perceived improvements (e.g., because the child was no longer signaling to parents they were awake). Conversely, it is possible that the child's sleep did improve, however improvements were not adequately captured by actigraphy due to its reliance on measuring restlessness. Thus, the results of actigraphy may need to be considered with caution among paediatric populations (Meltzer et al., 2012). It is therefore important to acknowledge that sleep diaries are a valuable source of information, and as change was identified in sleep diaries, the benefits of the interventions within majority of these studies should not be discouraged.

Finally, research has found that sleep problems and mental health challenges (e.g., depression and anxiety) in children with CHC are inter-related (Palermo & Kiska, 2005; Hysing et al., 2009; Mulrooney et al., 2008; Zeltzer et al., 2009; Lewandowski et al., 2011a; Pavlova et al., 2017). Additionally, research in children with CHC have reported an increased prevalence of sleep difficulties (Sadeh et al., 1998; Becker et al., 2004; Bursztein et al., 2006; Heng & Wirrell, 2006; Newman et al., 2006; Naqvi et al., 2008; Sivertsen et al., 2009; Lewandowski et al., 2011a; Tsipoura et al., 2018; Lang et al., 2021) and psychological difficulties (Lavigne & Faier-Routman, 1992; Bennet, 1994; Dantzer et al., 2003; LeBovidge et al., 2003; Smith et al., 2010; Reynolds & Helgeson, 2011; Moreira et al., 2013; Moreira et al., 2015; Quittner et al., 2016; Lewandowski et al., 2011a). Importantly, as sleep difficulties and psychological difficulties are common in these children, the findings of this review provide valuable information regarding how improving sleep may also benefit a child's

daytime behaviours, and mental health and wellbeing. Specifically, in the current review, sleep interventions were associated with improved daytime behaviours, quality of life and psychological outcomes such as depression and anxiety symptoms. Thus, these findings support the interrelationship between sleep and wellbeing, and the secondary benefits of sleep interventions in improving various domains of a person with a CHC life, which have the potential to further break negative cycles that maintain sleep difficulties.

Limitations and Directions for Future Research

Although these findings are encouraging, the results should be interpreted with caution due to multiple limitations. Firstly, because psychosocial sleep interventions are often multimodal, difficulties can emerge when attempting to factor out which components of the treatment are responsible for improvements (McLay et al., 2020). Because all of the studies included in this review utilised a combination of treatment approaches, with multifaceted delivery in some instances, it is not possible to draw conclusions on which elements were the most effective. In situations where clinicians are time and resource limited, it may not be possible to employ all the strategies used within these studies, and it may also be difficult for parents to implement multiple strategies for the same reasons. This is important as research has demonstrated that managing various CHC can be very time-consuming for parents (Melnyk et al., 2001; Cousino et al., 2013; Santer et al., 2014; Kish et al., 2018), which could then contribute to poor adherence for highly intensive sleep interventions. Future research should therefore consider designing interventions in a way that enables investigation of the relative contribution of different components to determine which components are necessary (e.g., staggering the introduction of treatment components).

It is also important that future research investigate which modality of treatment delivery is best suited for children with different CHC. Delivering interventions so that they are minimally sufficient to produce treatment gains, while not producing further treatment

burden is important for families with CHC who may already be experiencing significant illness burdens (Jowsey et al., 2012) and have limited financial resources (Druss et al., 2001). A majority of studies in this review delivered intervention face-to-face or via video sessions, while two studies utilised online programs (one using a combination of clinic visits and online modules [Sonney et al., 2020], and one being delivered fully by an online program [Zhou & Recklitis, 2020]). Online interventions can allow families to consume treatment information at their own pace (McLay et al., 2020), which may increase the utility and feasibility of interventions for families with high health needs. Law et al. (2018) found that the enrolment rate in their study was only 35%, possibly owing to accessibility issues related to the distance for families to travel to the research institute or time requirements. They suggested that delivering interventions online could help to improve accessibility, thus the use of telehealth delivered psychosocial sleep interventions for children with CHC may be an important area to explore in future research.

There were also many limitations identified by the authors of the included studies which may have compromised data reliability. These included; small sample sizes, the lack of diversity within participants and studies, and a lack of long-term follow up. This issue is further compounded by lack of replication of treatment effects across different CHC. Importantly, although this review was evaluating sleep interventions for children with CHC, the research obtained is not representative of all CHC diagnoses. The term CHC encompasses many different diagnoses, which affect a child physiologically, emotionally and behaviourally in unique ways. Thus it cannot be guaranteed that an intervention which improves sleep within one CHC will prove as effective in another. As the studies in the review only focused on some CHC, with only 2 – 3 studies for each condition, the research is limited to a small range of CHC, and may not be generalisable to other CHC (e.g., crohns disease, CF, heart disease) that were not included in this review. Furthermore, given the low

number of studies identified in this review, the evidence base for psychosocial sleep

interventions in children with CHC as a whole is yet to be determined. Thus, further research

is needed to replicate treatment effects and extend research across a wider range of CHC.

Relatedly, participants in the reviewed research were predominantly from the U.S

(8/13 studies) and Caucasian. The findings of this review are therefore not only limited in

terms of the range of CHC represented, but also the lack of cultural diversity. Being part of a

marginalised group is significantly associated with an increased risk of CHC, a greater

likelihood of high psychological distress and of illness- or injury-related absence from school

or work (Robards et al., 2020). Thus, it is important that research pays equal attention to

children of different ethnicities, social-economic status and health status.

There were also various limitations discussed within the studies in regard to

measurement, with authors citing a lack of objective measures (Bartlet & Beaumont, 1998;

Meltzer & Booster, 2016; Zhou & Recklitis, 2020); a variety of measures not being assessed

at multiple time points (Zhou & Recklitis, 2020); and a reliance on only one subjective report

(Meltzer & Booster, 2016; Sonney et al., 2020), which could have resulted in social

desirability biases. Overall these limitations affected research quality judgements. Future

studies can therefore build upon existing research through greater use of objective measures

of sleep and collection of IOA data.

Future assessments should also consider additional secondary outcome domains such

as emotional, behavioural, and physiological outcomes. Of the studies in the current review

that investigated non-sleep outcomes, it is important to acknowledge that the study by Law et

al. (2018) reported improvements in the frequency of headaches following the sleep

intervention, illustrating the potential relationship between illness symptoms and sleep.

Therefore, the potential mediating relationship between illness severity and a child's sleep

may be important to understand if similar improvements can be seen within other CHC.

Three studies (Palermo et al., 2016, 2017; Law et al., 2018; Tsai et al., 2020) within the review also discussed how the concurrent use of medications or other treatments during intervention may have may have impacted results. Therefore it was not possible to determine that improvements were solely attributable to the sleep intervention. This may be a common limitation to many of the studies within this review, as many children with CHC require medications and extensive treatment regiments to manage their symptoms (Fielding & Duff, 1999; So, 2013; Russo, 2022). It would be unethical to limit participants access to medications which benefit their overall health, therefore this cannot be controlled for. Law et al. (2018) did however propose that future studies may be able to tease apart the impacts medication have on treatment success through RCT's that compare medications on their own with the combination of sleep interventions.

Future studies should also consider the health needs within children with CHC, as well as investigate the use of sleep interventions across varying levels of symptom severity, especially as research has reported that worse physical health is associated with worse sleep in children with CHC (Lewandowski et al., 2011a). Health needs may also fluctuate for these children, therefore monitoring symptoms will be an important component of sleep interventions. Monitoring symptoms would ensure that interventions do not pose a threat to the health of the child, as well as providing a means to determine if improving sleep also improves other health-related outcomes for the child.

Although the current review focused on studies that included at least one child with a CHC, in four studies participants were mixed and included children with CHC as well as healthy children and/or children with neurodevelopmental disorders. Therefore, the results cannot be fully attributable to a population of solely CHC diagnoses. It is also possible that relevant studies may not have been identified within the search for evaluation, owing to the inclusion/exclusion criteria of this review. Additionally, it is possible that other studies not

identified in this review may have included children with CHC, however these studies may not have disclosed diagnoses, therefore not meeting the current inclusion criteria.

Lastly, as sleep problems are common among children with CHC, it is important to note that more may need to be done in terms of screening for, assessing and supporting families to prioritise sleep problems where they exist in children with CHC. This is important as sleep problems may be missed in many children or left untreated due to more complex and pressing symptoms taking priority, despite the importance of sleep for overall health.

Conclusions

Improving children's sleep not only positively impacts their emotional and social wellbeing, but also their physical health. This is especially paramount for children with CHC. Despite the need to address sleep problems in children with CHC, there is a lack of research investigating the effectiveness of psychosocial sleep interventions for children with CHC. Only 13 studies were identified within the current review. These studies varied in their methodological quality, with many limitations pertaining to the exploratory nature of the studies. The findings of the studies did however suggest that children with CHC who have sleep problems may benefit from psychosocial sleep interventions. The included studies utilised behavioural strategies, CBT-I and MBI, which resulted in 12/13 studies finding at least some improvements in sleep. Additionally, in the studies that assessed secondary outcomes, some improvements were seen in symptoms of depression and anxiety, HRQOL and daytime behaviours following the sleep intervention. These findings are in line with the literature exploring similar interventions within typically developing children and children with neurodevelopmental disorders. More research is however required which utilises high quality methodologies to extend and replicate treatment effects. Additionally, it is important that further studies continue to consider the significance of the specific CHC when intervention planning and explore how the sleep intervention may also improve physical

symptoms of the CHC. The current review therefore demonstrates that there is promising

evidence for the use of psychosocial sleep interventions within children with CHC, however

further methodologically rigorous research is required.

Chapter Three: Case Study

This chapter presents a case study examining the effectiveness of a behavioural sleep intervention for a child with CF and sleep problems. Following the results of Chapter Two, it was concluded that behavioural sleep interventions are likely to be effective in improving sleep difficulties in children with various CHC. Based on these findings, it was hypothesised that an individualised Functional Behaviour Assessment (FBA) informed sleep intervention, incorporating key elements utilised within the effective studies reviewed in Chapter Two, may effectively treat sleep problems in a child with CF. Additionally, because the findings of Chapter Two suggested that many of the typical sleep intervention strategies used for healthy children were effective for children with various CHC, it was assumed that such strategies could be safely used for a child with CF. The methodology for this case study was informed by the findings of Chapter Two in combination with the research discussed in Chapter One. The clinical implications of the findings, directions for future research, and the strengths of the conclusions of Chapters Two and Three are discussed in Chapter Four.

This study aimed to answer the following research questions:

1. Can a behavioural sleep intervention effectively reduce sleep problems in a child with CF?

2. Will any treatment effects be maintained at Short Term Follow Up (STFU)?

3. Will the selected sleep intervention be acceptable to the parents?

4. Will improving the child's sleep affect the child's daytime behaviour and Health Related Quality Of Life (HRQOL)?

5. Will improving the child's sleep affect the wellbeing of their parents?

Methodology

The Sleep Research Team

The reported case study is based on data collected as part of a wider sleep research programme led by a team at the University of Canterbury. As author of this thesis, I was part of this team. I completed the recruitment for participants, and the academic work, including the organisation and analysis of data, the post-treatment interview, and writing and editing the original drafts. A clinician on the team led the clinical work, with support from the sleep research team.

Ethics and Participant Consent

The wider study this case study was part of received ethical approval from the University of Canterbury Human Ethics Committee (HEC2018-48). The child's parents provided written informed consent. A copy of the consent and information forms given to the family are attached (see Appendix A and B).

Research Design

This research utilised an AB case study design with a STFU phase. Case studies retain the richness of individual-specific data and provide the opportunity for meaningful interpretation of the effects of treatment (Priday et al., 2017). This design also allowed for monitoring of the participant's response to treatment, so that adaptations could be made to the treatment plan if and when required.

Participants

Recruitment. An advertisement (study flyer) was sent to organisations throughout New Zealand who provide services to families with a child with CF (e.g., CF New Zealand). The advertisement included information about the study and how to become involved. Modified social media friendly advertisements were also promoted on social media via the

research team and CF New Zealand. Families who contacted the research team were provided with further information and screened for eligibility.

Screening and Confidentiality. An initial screening process was completed over the phone by a member of the research team for families interested in participating. The purpose of the screening call was to ascertain whether the research study was appropriate for families and whether they were eligible for inclusion in the study. During this call, the study aims, a basic procedural outline, and information on confidentiality were discussed.

Inclusion/Exclusion Criteria. Children were eligible for inclusion in this study if they meet the following criteria: (a) were between the ages of 2-18 years; (b) had a diagnosis of CF, as verified by a paediatrician; and (c) had sleep disturbance, as indicated by parent or self-report, in the form of difficulty with sleep onset, frequent and/or prolonged Night Waking (NW) and/or daytime sleepiness. Children were excluded if they: (a) were undergoing active medical treatment; and/or (b) had medical symptoms that were presently affecting their sleep and that were a contraindicator for behavioural intervention.

Participant Characteristics. One participant met criteria for inclusion in this study. Matthew (pseudonym) was a five-year-old New Zealand European boy with CF, who lived at home with his mother and father. Matthew's father contacted the research team after viewing the advertisement through social media. Matthew was pancreatic sufficient and had no other physical or developmental issues. Matthew was receiving ongoing treatment of Tobramycin, via a nebuliser, which is typically used to treat lung infections. This was administered twice per day, every second month, for 20 minutes, to keep any infection at a low level. Every morning, Matthew also completed Positive Expiratory Pressure (PEP) therapy. PEP therapy involves wearing a face mask that allows air to flow freely as a person breathes in, however resistance is created when air is expelled, resulting in making it more difficult to breathe out (Cystic Fibrosis Foundation, n.d.). This helps to build pressure in the lungs so that air can get

behind any mucus, thus moving mucus from the lung and airway walls (Cystic Fibrosis Foundation, n.d.). Matthew also completed 20 minutes of physiotherapy exercises for his lungs with his parents daily. This included his parents banging and massaging his chest. At the time intervention commenced, Matthew had recently begun primary school.

Setting. The intervention was implemented in the family home by Matthew's parents, with support from the clinician and wider study team. The clinical interview (described below) was completed via Zoom by the clinician, and subsequent contact with the family was maintained via email, phone (call or text), or Zoom.

Sleep Measures

Sleep data were obtained through the clinical interview, sleep diaries and the Children's Sleep Habits Questionnaire (CSHQ: Owens et al., 2000). These measures are described below. FBA was undertaken using the clinical interview and sleep diary data.

The Clinical Interview. An open-ended clinical interview was conducted via Zoom with the clinician (intern psychologist under the supervision of a registered psychologist) and Matthew's father. The clinical interview followed a format used by the Child and Family Psychology Programme and the Pukemanu Centre at the University of Canterbury. This comprehensive assessment was used for the purpose of assessing Matthew's sleep problems in the context of other relevant child and family factors. The format of the interview involved introductions and reiterating consent and confidentiality, then gathering information about his sleep problems, history of sleep problems (including any previous support or strategies tried) and parental goals for intervention. Information was also gathered about Matthew's family and home environment, developmental history, history of CF, and his strengths. For example, questions about Matthew's sleep problems included "tell me about the difficulties Matthew is having with his sleep", "how often does he wake in the night?", "how do you respond when he wakes?", "has his sleep gotten worse or better over time?", and "when did you first notice

Matthew having difficulties with sleep?". The use of sleep measures (e.g., sleep diaries), were also discussed, including guidance on how to use them.

Sleep Diaries. Parent-reported sleep diaries were completed on a Word document each night during baseline, intervention and STFU phases, and then emailed to the clinician weekly. Sleep diaries included questions about night-time sleep, including: (a) the setting where Matthew fell asleep; (b) who put him to bed; (c) the time he was put to bed; (d) the frequency of any curtain calls (see definition under Dependent Variables below), his behaviour during each curtain call and his parents' response to his behaviour; (e) his parents best estimate of the time he fell asleep; (f) the time his bedroom door was closed; (g) the time and duration of any NW; (h) his behaviour during NW; (i) his parent's responses to his NW behaviour; and (j) what time Matthew woke in the morning. Daytime sleep was not recorded as Matthew did not sleep during the day. Parents were also able to note any other variables that they thought may have affected Matthew's sleep (e.g., becoming unwell or intense daytime activities) on the sleep diaries. A copy of the standard parent-report sleep diary is attached (see Appendix C).

Matthew's parents were asked to complete sleep diaries within 30 minutes of him waking in the morning, or at any time when he woke in the night. Upon receiving the sleep diaries each week, data were graphed and visually analysed (see Data Analysis below), which enabled Matthew's sleep to be closely monitored by the research team throughout all study phases.

The Children's Sleep Habits Questionnaire. The CSHQ is a 33-item parent report questionnaire used to categorise children's sleep problems, including bedtime resistance, sleep onset delay, NW, sleep-related anxiety, sleep disordered breathing and daytime sleepiness. Parents rate the frequency of specific sleep behaviours over the previous week using a three-point Likert scale (usually = 5 – 7 nights per week; sometimes = 2 – 4 nights

per week; or rarely = 0 – 1 night per week). Questions also ask whether the sleep behaviours are considered problematic for the family (e.g., next to the frequency Likert scale, parents are instructed to circle "yes", "no", or "not applicable" regarding the same behaviour).

The CSHQ provides an overall total sleep disturbance score, with higher scores indicative of poorer sleep (the total score clinical cut-off is ≥41), as well as eight subscale scores relating to specific sleep disturbances commonly experienced by children: Bedtime Resistance, Sleep Onset Delay, Sleep Duration, Sleep Anxiety, NW, Parasomnias, Sleep Disordered Breathing, and Daytime Sleepiness. The CSHQ takes approximately 15 minutes for parents to complete.

The CSHQ has been found to have good psychometric properties (Hodge et al., 2012; Hoffman et al., 2006). Owens et al. (2000) reported that the CSHQ has adequate internal consistency within a community sample (α = .68), as well as in a clinical sample (α = .78), and acceptable test-retest reliability (ranging from .62 - .79). Owens et al. (2000) also reported the CSHQ is able to distinguish between control and clinical groups, with a sensitivity of 0.80 and specificity of 0.72. The CSHQ has been used widely to study sleep in typically developing children, and has also been used within children with CHC (e.g., Tsai et al., 2020; Zupanec et al., 2017). One study by Lewandowski et al. (2011b) reviewed parent and child-report paediatric sleep measures according to the criteria developed by the American Psychological Association (APA) Division 54 Evidence-Based Assessment (EBA) Task Force, and reported the CSHQ as "well-established". This meant the psychometric properties of the CSHQ were considered sound (Lewandowski et al., 2011b). Within the present study, Matthew's father completed the CSHQ.

Secondary Outcome Measures – Child

The Child Behavior Checklist 1.5-5 years (CBCL; Achenbach & Rescorla, 2000) and the Pediatric Quality of Life Inventory (PedsQL; Varni, 1999) were administered pre- and

post-treatment to assess change in Matthew's emotions and behaviour, and quality of life, respectively. Matthew's father completed both the CBCL and PedsQL.

The Child Behavior Checklist. The CBCL (1.5 – 5 years) is a 100-item standardised parent report questionnaire that assesses internalising, externalising and problem behaviours in children aged 18 months to five years. Parents report the frequency of behaviours in their child on a three-point Likert scale: (0 = not true; 1 = somewhat or sometimes true; or 2 = very true or often true). The CBCL includes eight subscales (Emotionally Reactive, Anxious/Depressed, Somatic Complaints, Withdrawn, Sleep Problems, Attention Problems, Aggressive Behaviour, and Other Problems). The subscales are combined to form total Internalising Problems, Externalising Problems and Total Problems composite scores. The composite scores are then summed and converted into standardised T scores, with higher T scores indicating greater emotional and behavioural difficulties. T scores are classed as falling within a normal, borderline, or clinical range.

The CBCL is considered reliable with high validity (Iyanova et al., 2010), and is able to distinguish between control and clinical groups, with a sensitivity of 0.85 and specificity of 0.90 (Achenbach, 2001). Additionally, test-retest reliability and internal consistency have been reported as high. Albores-Gallo et al. (2007) reported test re-test reliability as ≥ 0.95 for the internalising, externalising, and total problems subscales, and internal consistency scores of α= 0.89-0.95 for the internalising, externalising and total problems subscales.

The Pediatric Quality of Life Inventory. The PedsQL is a 23-item measure of HRQOL for children. For the present study, the PedsQL parent-report for young children (ages 5 – 7 years) generic core scales were utilised in accordance with the scaling and scoring guidelines created by Varni et al. (1999). The PedsQL measures four dimensions of health including physical, emotional, social and school functioning. Respondents are instructed to consider the magnitude a problem has been for the child over the past month, and rate items

using a 5-point Likert scale (0 = never a problem; 1 = almost never a problem; 2 = sometimes a problem; 3 = often a problem, 4 = almost always a problem). The PedsQL items are reverse scored and transformed to a 0 – 100 scale (0 ["Never")] = 100; 1 ["Almost Never"] = 75, 2 ("Sometimes") = 50, 3 ("Often") = 25, 4 ("Almost Always") = 0), so that higher scores indicate better HRQOL. For each of the four scales, scores are then computed to create a sum of the items over the number of items answered (to account for missing data). This results in four subscale scores and a total score, again with higher scores indicating better HRQOL.

The PedsQL has been demonstrated to have good to excellent psychometric properties. For example, Varni et al. (2001) reported the PedsWL to have an internal consistency total score of 0.88 for child-report, and 0.90 for parent-report within a paediatric sample of 963 healthy children and 1,629 parents. The PedsQL has also been reported to have adequate divergent validity as it can differentiate between healthy children and those with CHC (Varni et al., 2003).

Secondary Outcome Measures – Parents

The Pittsburgh Sleep Quality Index (PSQI; Buysse et al., 1989), Depression Anxiety and Stress Scales – 21 (DASS-21; Lovibond & Lovibond, 1995) and Relationship Quality Index (RQI; Norton, 1983) were administered pre- and post-treatment to assess change in parental sleep quality, well-being and relationship quality, respectively. Both of Matthew's parents completed the RQI at pre- and post-intervention, however only Matthew's father completed the DASS-21 and PSQI at both time points.

The Pittsburgh Sleep Quality Index. The PSQI is a 19-item self-report questionnaire designed to measure the quality and patterns of sleep in adults (Buysse et al., 1989). Participants are asked about their usual sleep habits over the past month. The PSQI produces a global sleep quality score, with higher scores denoting more impaired sleep, and seven component scores related to sleep quality, sleep latency, sleep duration, habitual sleep

efficiency, sleep disturbance, use of sleeping medications, and daytime dysfunction over the last month. The PSQI uses varying response categories, including instructing respondents to record their usual bed and wake times, the number of actual hours spent asleep, the number of minutes taken to fall asleep, as well as Likert-scale responses for how often items have affected them in the past month (Buysse et al., 1989).

Buysse et al. (1989) reported the PSQI has good psychometric properties, with a sensitivity of 89.6% and specificity of 86.5%, as well as an internal reliability of .83, and test-retest reliability of .85.

The Depression Anxiety and Stress Scales –21. The DASS-21 is a 21-item retrospective self-report scale designed to measure features of depression, anxiety and stress in adults. Results indicate the presence of psychological distress (Henry & Crawford, 2005). Respondent are asked to consider how much a statement has applied to them over the past week using a four-point Likert scale (Never [did not apply to me at all)]; Sometimes [applied to me to some degree, or some of the time]; Often [Applied to me a considerable degree, or a good part of the time]; Almost Always [Applied to me very much, or most of the time]). The sum of seven items for each of the three axes provides a score, which determines the severity of symptoms (Normal, Mild, Moderate, Severe, or Extremely Severe).

The DASS-21 has been used widely to assess adult well-being in both research and clinical settings. Henry and Crawford (2005) reported the DASS-21 has adequate reliability for the subscales (α=.82 - .90), and good convergent and discriminative validity. Additionally, Gloster et al. (2008) assessed the DASS-21 and found it had good internal consistency, convergent validity, and discriminative validity, especially with the depression scale.

The Relationship Quality Index. The RQI is a six-item self- report questionnaire designed to assess the quality of relationships in married and cohabiting couples.

Respondents individually rate how much they agree with five statements. Each statement evaluates respondents' level of satisfaction in different aspects of their relationship using a 7-point Likert scale (1= very strongly disagree, through to 7= very strongly agree). Respondents also provide an overall rating of their level of happiness in the relationship on a 10-point Likert scale (1 ["unhappy"]) to 10 ["perfectly happy"]). The scores are then summed to indicate overall relationship satisfaction, with higher scores indicating better subjective relationship quality, and scores ≤ 29 indicating relationship distress.

Treatment Acceptability

Treatment acceptability was measured using a post-treatment interview and the treatment Acceptability Rating Form – Revised (Reimers & Wacker, 1992). Matthew's father completed both.

The Post-Treatment Interview. In order to gain an understanding of the parents' perspective of the intervention process, an interview was conducted immediately following treatment by the thesis author. Only Matthew's father was present for the interview. During the interview, he was asked how he felt about the intervention and the overall process, as well as his perspectives on whether treatment was effective and his attributions about treatment effects. He was also asked whether the process had any impact on the family, for them as parents or on other areas of Matthew's behaviour or development (e.g., on his CF symptoms). The interview also provided an opportunity for feedback on how the assessment and treatment process could be improved.

The Treatment Acceptability Rating Form – Revised. The TARF-R is a 20-item parent-report questionnaire used to determine the acceptability ratings of interventions for children in natural settings (Reimers & Wacker, 1992). It is administered post-intervention to obtain perspectives in regard to eight subscales: Reasonableness, Effectiveness, Side Effects, Disruptive/Time Consuming, Cost, Willingness, Problem Severity, and Understanding (of the

treatment process). Respondents are asked to consider how appropriate, effective and fair they deem treatment to be for 17 items, and the remaining three items assess the respondent's understanding of the intervention, and their perception of the severity of their child's behaviours. A 7-point Likert scale (e.g., with ratings of "not at all effective" to "very effective" etc) is used to determine scores, and items are summed to give a total acceptability score. Higher scores indicate higher acceptability of treatment. The TARF-R has been reported to have good reliability ($\alpha = .92$) and clinical utility (Finn & Sladeczek, 2001; Reimers & Wacker, 1992).

Dependent Variables

Key dependent variables included awake, asleep, co-sleeping, parental presence, curtain calls, Sleep Onset Latency (SOL), NW, Total Sleep Time (TST) and Early Morning Waking (EMW). Parent-report data from sleep diaries were used to measure the dependent variables which are defined below.

Awake. Awake was defined as Matthew having his eyes open and/or engaging in purposeful gross motor movements and/or vocalisations that were indicative of wakefulness (e.g., sitting up, looking around, calling out).

Asleep. Asleep was defined as Matthew laying still in his bed with his eyes closed and not making purposeful gross motor movements or vocalisations associated with wakefulness.

Co-Sleeping. Co-sleeping was defined as Matthew lying in the same bed as his parents, or his parents lying down on a mattress in his bedroom for any duration during the night including settling to sleep or following a NW. This included co-sleeping initiated by Matthew (i.e., where Matthew requested his parents to lie in his room), and parent-initiated co-sleeping (i.e., where Matthew's parents lay on the mattress in his room without his request).

Parental Presence. Parental presence was defined as either of Matthew's parents being in physical or visual proximity to Matthew during sleep onset for longer than five minutes. Parental presence included his parents being in the same room (e.g., sitting inside his room until he fell asleep) and co-sleeping.

Curtain Calls. Curtain calls were defined as any bids for parental attention after Matthew was put to bed, including if he remained in bed and called out to his parents or if he got out of bed. Each occurrence of one of these behaviours was recorded as a curtain call.

Sleep Onset Latency. SOL was defined as the duration of time (in minutes) that it took for Matthew to fall asleep. SOL was calculated from the time his parents reported they left his room, until their estimate of when he fell asleep.

Night Waking. NW were defined as any arousal in which Matthew signalled to his parents that he was awake (e.g., called out to them, or got out of bed) between the time he first fell asleep until the time he got up in the morning. Matthew's parents estimated the duration of his NW from the start to end (i.e., when he resumed sleep).

Total Sleep Time. TST was defined as the total duration of time (in minutes) that Matthew slept each night. TST was calculated from the time his parents reported he fell asleep to the time he woke for the day, minus the duration of any NW.

Early Morning Waking. EMW were considered any occurrence where Matthew awoke before his scheduled wake time and did not return to sleep (e.g., if he awoke at 5.45am but fell back to sleep and woke again at 6.30am this was considered a NW and his wake time would be recorded as 6.30am).

Presenting Sleep Problems

During the clinical interview Matthew's father reported that Matthew woke at least once most nights of the week between 11.30pm and 4.00 a.m. Following a NW, Matthew would enter his parents' bedroom and wake them. His parents typically responded by

returning him back to his room and placing a mattress on the floor, where either parent would then lie next to him. Matthew's mother would wait for him to fall asleep (usually within 20 mins) and then leave the room, whereas his father would often fall asleep on the mattress and remain there for the duration of the night. Matthew's father reported that approximately once per fortnight Matthew may remain awake for up to two hours during NW.

Matthew had a consistent bedtime routine that involved him having a bath/shower, completing bed-preparation activities (e.g., brushing teeth, pyjamas) and reading stories for 20 – 30 minutes at the consistent time of 7.00pm. Following the stories, his parents would turn off the light and leave the room, leaving him to fall asleep independently (i.e., without a parent present) by 7.30pm. Matthew had a special blanket which he used as a comfort item. He typically slept for 10 – 11 hours and woke independently between 5.30am – 6.00am. Matthew did not nap during the day. Matthew's parents were concerned by NWs and the co-sleeping that resulted in order for him to resume sleep. They also preferred him to remain in bed for longer before waking for the day (i.e., not wake before 5.30am). Matthew's parents' goals were for him to sleep through the night independently without waking until an appropriate time in the morning.

History of Sleep Disturbance

According to Matthew's father, Matthew had experienced poor sleep owing to NW and EMW since he was young. To support his sleep, his parents were using a sleep clock. Sleep clocks are a type of visual support to provide discriminative stimuli for bed and wake times (i.e., a visual representation of day/night to children's understanding of when to stay in bed and when it is acceptable to get up). Matthew's sleep clock changed from red (signalling sleep time) to green (signalling awake time) when it was an acceptable time for Matthew to get up from his bed. Matthew's father felt the clock was ineffective because even though he reported that Matthew understood what the colours meant, Matthew continued to get out of

bed during the night and before it displayed that it was time to get up in the morning. Matthew's parents had also attempted a reward system in the past where Matthew received a toy when he slept in his own bed for three nights in a row, however, this did not result in any beneficial change as he continued to require parental presence during NW.

Functional Behaviour Assessment

FBA indicated that Matthew's NW were primarily perpetuated by access to parent attention. Co-sleeping reinforced Matthew's behaviour of waking and seeking his parents' attention during the night. This likely resulted in a 'behaviour trap' where his parents were co-sleeping to get him to return to sleep, however this unfortunately increased the likelihood he would continue to wake over time and require their presence. Although Matthew was able to self-settle independently at the start of the night, a history of co-sleeping in response to NW meant that he may have experienced an inability to re-settle, thereby relying on parental presence to resume sleep. His behaviour of getting out of bed and/or calling out to his parents when awake therefore actively interfered with his ability to resettle quietly and independently.

In addition, Matthew may have experienced a lack of physiological sleep pressure to sustain sleep until 6.00 am. Insufficient sleep pressure can result in difficulties initiating and maintaining sleep (Jenni & LeBourgeois, 2006). Matthew's typical TST was approximately 11 hours which is within the recommended duration of 10 – 13 hours for children between 3 – 5 years of age (Hirshkowitz et al., 2015). However, his bedtime of 7.30pm may have been too early as it meant he had met his total sleep needs (of between 10 – 13 hours TST) before 6.00am, possibly resulting in early wakes and/or fragmented sleep.

Study Phases

The phases of the study were assessment (including FBA) and treatment planning, baseline, intervention with three subphases, maintenance, and STFU. These are explained below.

Assessment and Treatment Planning. The assessment phase involved conducting the clinical interview and FBA; this information was used to inform Matthew's treatment plan and was formulated by the sleep team. Treatment components were also individualised to Matthew's family's needs, strengths, and goals. Prior to commencing intervention, the clinician provided information and discussed the components of the treatment plan with the family. The CSHQ, CBCL, PedsQL, PSQI, DASS-21, and RQI were administered at this stage (i.e., pre-intervention assessment).

Baseline. Following the initial assessment procedures, the family completed a baseline phase where sleep diaries were collected for 19 nights. This was initially expected to last fourteen nights, however was extended due to Matthew becoming unwell. Matthew's parents were advised to maintain his normal sleep habits and routines, as well as their typical responses to his sleep behaviour so that information would accurately reflect Matthew's existing sleep patterns.

Intervention. Intervention occurred immediately after baseline, provided a stable baseline was evident. This ensured that any changes in sleep behaviours post-intervention could be attributed to the introduction of the intervention, rather than natural variations in Matthew's behaviour (Blampied, 2013; Kazdin, 1981). Intervention consisted of three subphases, lasting a total of 91 nights. Treatment components were subsequentially introduced in response to data collected from Matthew's sleep diaries. The implementation of new subphases occurred if treatment reductions of the magnitude intended were not yet demonstrated. Subphases were ordered according to the principles of minimal sufficiency

(i.e., less restrictive and time-consuming; van Deurs et al., 2021), whereby antecedent based factors were first addressed before the intervention moved to addressing consequence-based factors.

Regular contact was maintained between the clinician and Matthew's father throughout the intervention, which enabled the clinician to track Matthew's progress and to support the family with adhering to the treatment plan. Intervention proceeded until the sleep problems had reduced, and Matthew's parents and the clinician were satisfied that the family's goals had been met.

Intervention Subphase 1. The first subphase of intervention lasted 17 nights and involved modifications to Matthew's sleep/wake schedule. Matthew's regular bedtime was moved from 7.00pm to 7.30pm (i.e., his parents began reading him his bedtime stories at 7.30pm) to increase sleep pressure and the likelihood that Matthew would sustain sleep until his wake time. Matthew's parents were encouraged to maintain a consistent and calming bedtime routine during this subphase, and his wake time was kept at 5.45am

Two additional changes were made on Day 11 and Day 17, where Matthew's wake time was moved to 6.00 a.m. and 6.15 a.m., respectively, to accommodate family needs during the school holidays and his subsequent return to school.

Intervention Subphase 2. Intervention Subphase 2 (nights 18 – 50) involved the introduction of a personalised social story and reinforcement system.

Social Story. A personalised social 'sleep' story was developed for Matthew following the convention of Gray (2010). Photos of Matthew were collected from his parents and were accompanied with text written in brief, straight-forward first-person language (e.g., "I am learning to sleep through the night by myself"). The sleep story depicted his new bedtime routine, appropriate sleep behaviours (e.g., staying in his bed all night) and what he should do if he awoke in the night (e.g., cuddle his blanket and go back to sleep).

The story also instructed Matthew to follow the colours on his sleep clock if he woke during the night. For example, the story read; "If I wake in the night and my clock is red, I will stay in my bed" (paired with a photo of his sleep clock showing red), "I will cuddle my blanket and go back to sleep" (paired with a photo of Matthew cuddling his special blanket), and "When my clock is green, it is morning, and I can go to see Mum and Dad" (paired with a photo of his clock showing green). The sleep story ended with him receiving a reward for staying in his bed all night. The story was printed and sent to the family who were instructed to read this every night as part of their bedtime routine.

Reinforcement. A reward system was established whereby Matthew received a small toy in the morning if he was able to stay in his bed all night without going to his parents' bedroom. Matthew was provided with this reward immediately upon waking in the morning. The reward system was explained to Matthew verbally by his parents and reflected in the social story.

Intervention Subphase 3. Intervention Subphase 3 (was introduced on night 51 and lasted 41 nights) focused on teaching Matthew to re-settle to sleep independently if he awoke during the night. Prior to this subphase, his parents had continued to lie on the mattress in his bedroom in response to NW. During Subphase 3, Matthew's parents were instructed to avoid sitting/sleeping in Matthew's room when he woke in the night. Instead, they were instructed to provide assistance if needed (e.g., water or pain relief), and if he entered their room during the night, they would then return him to his bed with minimal interactions. Once back in his bed, his parents would say "I'm just going to go to the toilet" (or another "excuse me for a moment" type statement) and leave his room to give him opportunity to fall asleep independently. On the occasions when they returned to his room to check on him (within 5 mins), they reported he would be asleep, and they would then return to their room for the night.

Maintenance. Following the intervention phase, Matthew's parents entered a maintenance phase where there was no regular contact with the research team and data collection temporarily ceased. During this period, Matthew's parents were asked to continue using strategies as they did during intervention in order to consolidate the learning gained through treatment. The CSHQ, CBCL, PedsQL, PSQI, DASS-21, and RQI were administered (i.e., post-intervention assessment) along with the post-treatment interview and TARF-R.

Short Term Follow-up. Sleep diary data were collected for a one-week period at six weeks post-intervention to measure the maintenance of treatment effects over time.

Data Analysis

Data obtained through sleep diaries during baseline, intervention and STFU phases were graphed according to the targeted sleep variables for Matthew. The primary means of data analysis was systematic visual inspection of the graphed data, including assessment of changes in the trend, level and stability of data between subphases (Cohen et al., 2014). A relationship between the independent and dependent variables was considered present when a clear, consistent change in target sleep behaviours was demonstrated following the intervention (Vannest & Ninci, 2014). Visual analysis is a useful way to assess treatment outcomes, and has been used widely within case studies as it enables researchers to identify is behaviour changes are attributable to an intervention or not (Blampied, 2013; Hanley et al., 2003).

Changes in the scores of sleep (the CSHQ) and secondary child (the CBCL and PedsQL) and parent (the PSQI, DASS-21 and RQI) psychometric measures were assessed by comparing pre- and post-treatment scores, which also included consideration of whether scores moved out of their respective clinical ranges. For all measures except the PedsQL and RQI an improvement was indicated by a reduction in scores, while an increase in scores on

the PedsQL and RQI indicated improvement. The results of psychometric measures are reported in Tables 5 – 11.

Results

Sleep Intervention Outcomes

The frequency of curtain calls, duration of SOL (mins), frequency and total duration (mins) of NWs, whether co-sleeping occurred, and frequency and total duration (mins) of EMW are represented in Figures 2 – 7, respectively. Data for each dependent variable is presented across the baseline, intervention and STFU phases.

Curtain Calls. Curtain calls were not identified as a parent-reported problem; however, they were monitored to gain an overall understanding of Matthew's sleep, including his initial settling at the start of the night. The frequency of curtain calls is presented in Figure 2. During baseline, curtain calls ranged from 0 – 3 (M = 0.63). During intervention, curtain calls ranged from 0 – 1 (M = 0.13) in Subphase 1, from 0 – 3 (M = 0.35) in Subphase 2, and 0 – 5 (M = 0.46) in Subphase 3; meaning there was no improvement from baseline.

Further analysis of sleep diary data showed that there were only seven nights throughout intervention where more than one curtain call occurred. Three of these nights correlated with Matthew being sick and requiring pain relief, stopping his nebulising treatments that day (completed month on/off), or starting the new month of nebulising the same day. Further, 3/7 curtain calls only required minimal verbal encouragement from outside of Matthew's room for him to resume sleep. On day 107 when the frequency of curtain calls peaked (N = 5), Matthew required parental support due to complaining of being hot and thirsty. A sustained period of zero curtain calls per night occurred for 10 nights during intervention Subphase 1 (nights 29 – 38), and again for 12 nights during Subphase 3 (nights 93 – 104). This represents an improvement from baseline where the number of consecutive nights with zero curtain calls was four nights.

During STFU, the frequency of curtain calls ranged from 0 – 2 (M = 0.43), occurring on 2/7 nights. On the nights that curtain calls occurred, Matthew only required minimal encouragement from outside of his room one night, and a drink of water the other.

Figure 2

Frequency of Curtain Calls across Baseline, Intervention and Follow-up Phases
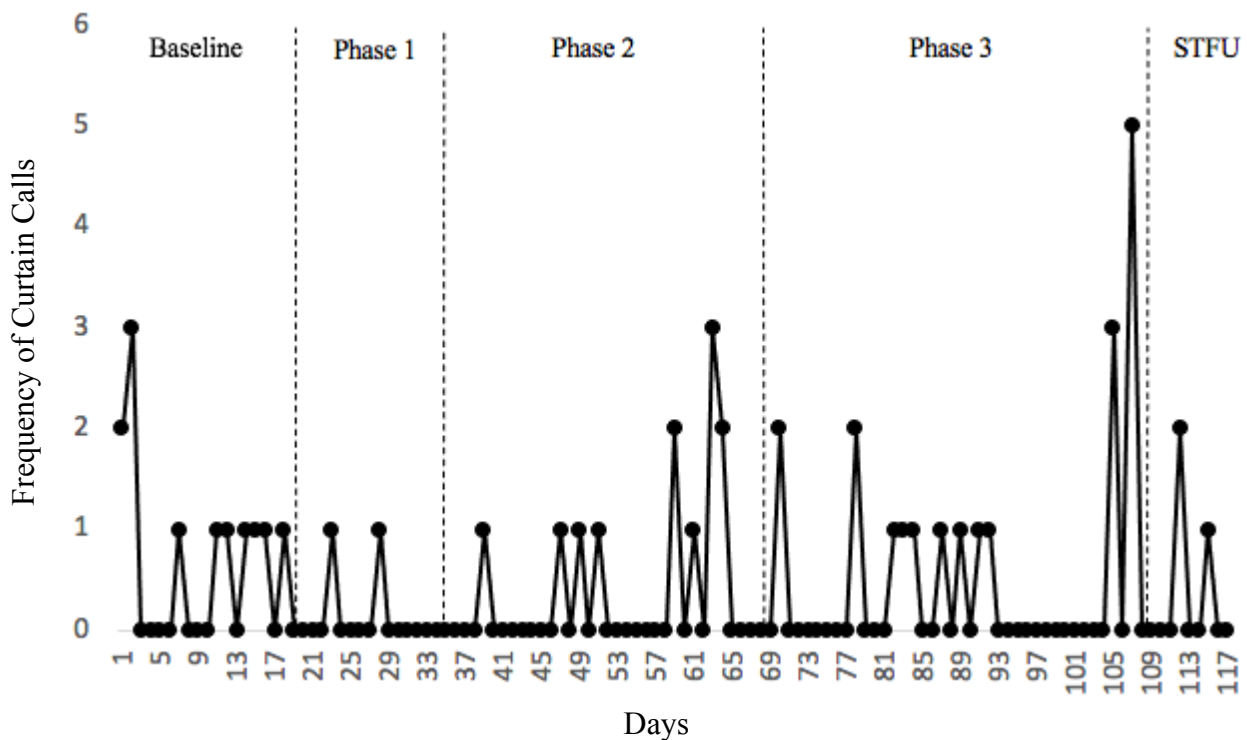
Sleep Onset Latency. Although SOL was not identified by Matthew's parents as a problem, it was monitored as part of his overall sleep picture. The duration of SOL (mins) is shown in Figure 3. During baseline, Matthew's SOL varied from 5 – 20 minutes (*M* = 9 mins). No treatment effects were evident during intervention, as his SOL ranged from 5 – 8 minutes (*M* = 5 mins), 5 – 35 minutes (*M* = 7 mins) and 5 – 40 minutes (*M* = 7 mins) during Subphases 1, 2 and 3, respectively. The longer SOL of 35 and 40 minutes on nights 59 and 107 (Subphases 2 and 3), respectively, were reported to be owing to sickness and a high bedroom temperature. During STFU, SOL remained within 5 – 10 minutes on 6/7 nights,

however on one night SOL reached 35 minutes (coinciding with two curtain calls) for reasons that were not explained by his parents.

Figure 3

Sleep Onset Latency across Baseline, Intervention and Follow-up Phases
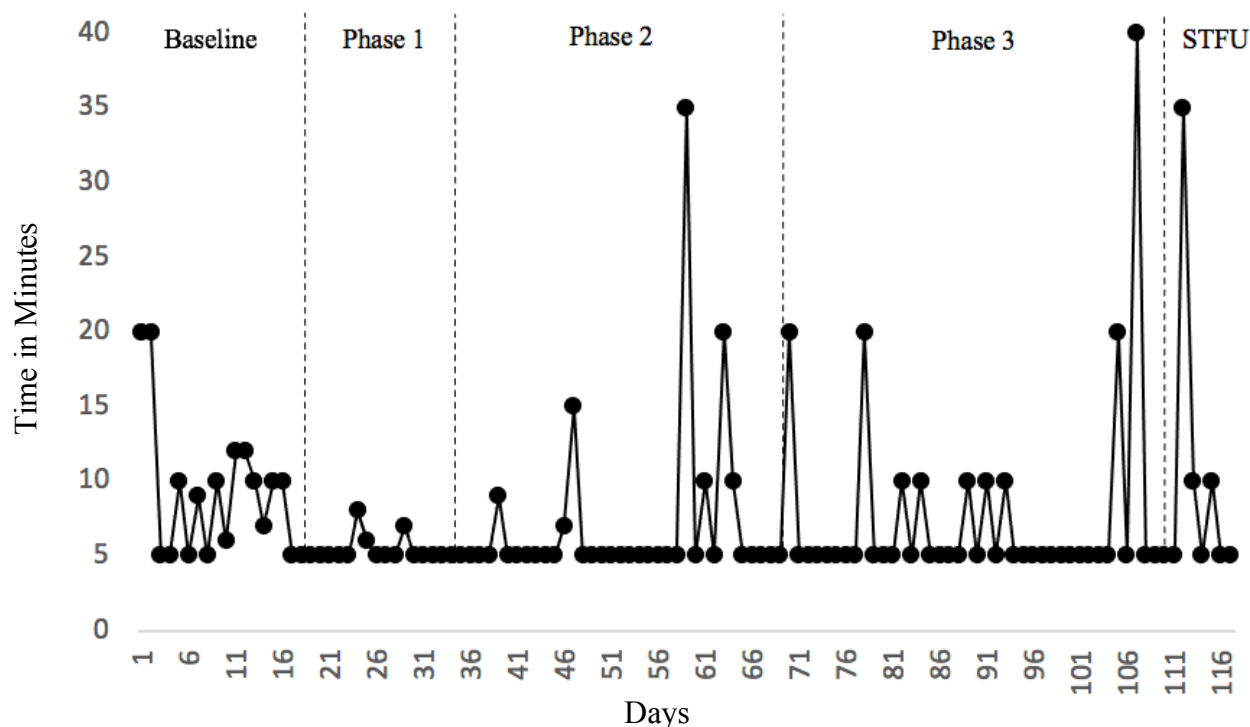
Frequency of Night Waking. The frequency of NW is presented in Figure 4. During baseline, NW occurred 0 – 4 times per night ($M = 1.26$) on 15/19 nights (79%). For every first waking, Matthew entered his parents' bedroom, and for subsequent wakings if his parents were not already co-sleeping with him, he would again go into their bedroom. In intervention Subphase 1 with the implementation of sleep/wake rescheduling Matthew's NW decreased slightly with NW occurring on 11/16 nights (69%), ranging from 0 – 5 ($M = 0.94$). For 10 of these nights, he woke once and entered his parents' bedroom. On night 26 when he woke five times, he was reported to be sick and required pain relief.

During Subphase 2, NW again reduced slightly, occurring on 22/34 nights (65%), and ranging from 0 – 4 ($M = 0.85$) in frequency. On three nights Matthew called out from his bed

to his parents, and for the other 19 nights he went into their room. There were 17 nights where only one NW occurred. The frequency of nights with no NW also increased slightly with Matthew experiencing five consecutive nights with no NW (nights 46 – 50), compared to baseline where three consecutive NW-free nights occurred. It is noteworthy that Matthew became sick multiple times during this phase and required pain relief (nights 47 – 49 and 53 – 59).

During Subphase 3, NW further decreased compared to baseline, occurring on 16/41 nights (39%), ranging from 0 – 4 per night ($M = 0.51$). On 2/3 instances where more than one NW per night occurred Matthew required pain relief. For 7/16 of the nights where NW occurred, Matthew called out from his bedroom, and on the remaining 9/16 he went to his parent's room after waking for the first time in the night. On the first night of Subphase 3, when Matthew awoke, his parents took him back to his room and reminded him of the plan to fall asleep independently. The frequency of nights with no NW also continued to increase during this subphase, with Matthew experiencing eight consecutive nights with no NW (nights 77 – 84).

During STFU, NW further decreased and occurred on 2/7 nights (29%) ranging from 0 – 2 per night ($M = 0.43$). This indicates that the reduction of NW during intervention compared to baseline was maintained over time. Anecdotally, Matthew's father also reported that he only experienced three NW during the 40-day maintenance period, suggesting that intervention effects were maintained during this time.

Figure 4

Frequency of Night Wakings across Baseline, Intervention and Follow-up Phases
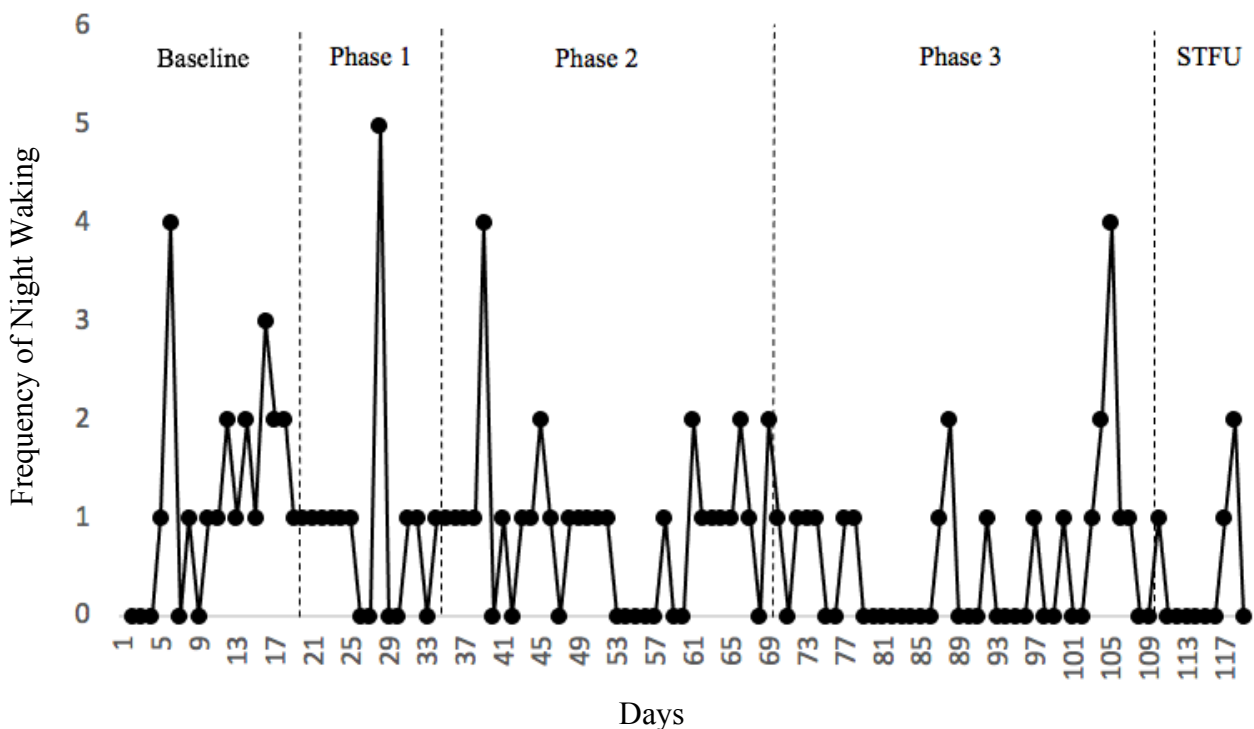
Total Duration of Night Waking. The total duration (mins) of NW is presented in Figure 5. During baseline, the total duration of NW per night ranged from 5 – 60 minutes (*M* = 18 mins) and there were four nights where singular NW lasted longer than 30 minutes (nights 11, 14, 18, and 19). During Subphase 1, the total duration of NW decreased compared to baseline, ranging from 5 – 10 minutes (*M* = 9 mins). On night 26 where the total duration was 50 minutes, five separate NW each lasting 10 minutes occurred. In Subphase 2, the total duration of NWs remained stable compared to Subphase 1, ranging from 5 – 20 minutes (*M* = 9 mins). In Subphase 3, the total duration of NWs remained stable compared to the previous subphases ranging from 5 – 20 minutes (*M* = 9 mins). Finally, during STFU, the total duration of NW ranged from 5 – 15 minutes (*M* = 10 mins). Overall, this showed a sustained improvement compared to baseline.

Figure 5

Total Duration of Night Wakings across Baseline, Intervention and Follow-up Phases
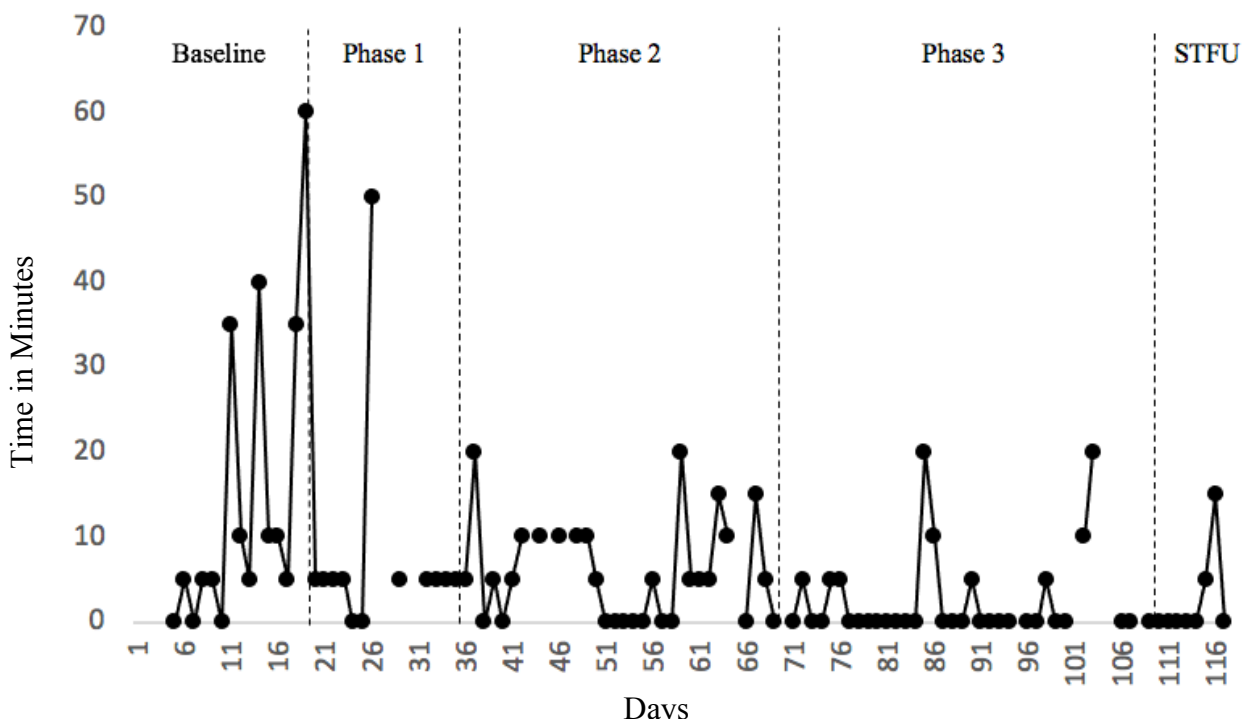
Parental Presence During Night Waking. Figure 6 shows the ways that Matthew's parents responded to NW. *Parent presence in child's room* was defined as any interaction which lasted > 5 minutes and included Matthew's parents either sitting on the bed or lying on the mattress until he fell asleep, or for the duration of the night (i.e., co-sleeping). *Child slept in parents' room* was defined as Matthew spending > 5 minutes sitting or sleeping in his parents' room (i.e., co-sleeping). *Redirected to bed* refers to instances where his parents returned him to bed and then left him to fall asleep independently. Finally, *no parental intervention required* was defined as any night where Matthew's parents were not awoken by him, so no parental intervention was required. This graph does not capture EMW (i.e., wakings that occurred before Matthew's scheduled wake time where he did not fall back to sleep); EMW data is presented in Figure 7.

During baseline, Matthew co-slept with his parents following 14/15 (93%) of the NWs that occurred. This included Matthew's parents sleeping in his bedroom following

11/15 NWs and Matthew sleeping in his parents' room following 3/15 NWs. This is captured in Figure 6 as "Parent presence in child's room" and "Child slept in parent's room", respectively. There was only one night where following a NW Matthew was provided water and then fell asleep while his father had left the room momentarily (i.e., redirected to bed). During baseline, the longest period of consecutive nights where parental presence was required (either in Matthew's room or his parents' room) was nine nights (between nights 8-16), and the longest period of consecutive nights where no parental intervention was required was two nights (nights 1 and 2).

Following the implementation of Subphase 1 (sleep/wake schedule changes), Matthew continued to experience NW, however at a slightly reduced frequency, and parental presence in his room following a NW occurred on 11/11 (100%) of the nights where NW occurred. Therefore, there was no reduction in parental presence following NW compared to baseline.

With the implementation of Subphase 2 the frequency of nights where parental presence was required (either in Matthew's room or his parents' room) decreased very slightly to 21/22 (95%), with Matthew requiring parental presence in his room to fall asleep for 19 of these, and Matthew sleeping in his parents' room for the remaining two. There was only one occurrence where redirection to bed was sufficient (night 68) and during this instance Matthew stayed in bed and appeared to be too hot, so his mother provided assistance and then left the room.

During Subphase 3 Matthew's parents were encouraged to redirect him to bed, unless support was required for health or safety reasons. His parents successfully redirected him to bed following 8/16 NWs (50%), which resulted in him sleeping independently (i.e., without parental presence) for the remainder of the night. Parental presence in response to NW occurred for the remaining 8/16 NWs (50%). This demonstrated a significant improvement

from baseline. Prior to this phase, the longest period of consecutive nights where no parental presence was required was five nights during Subphase 2 (nights 51 – 55), however during Subphase 3 there were 15 consecutive nights (nights 70 – 84) where no parental presence (either in his room or his parents' room) was required. Although parental presence continued to occur intermittently during Subphase 3, Matthew's parents reported they were satisfied that the frequency of nights where co-sleeping occurred had decreased.

During STFU, Matthew's parents continued to limit their parental presence during NW. On the two nights where he woke, his parents returned him to bed then left the room. During the subsequent wake on the second night, his mother remained in the room for 10 minutes, however it was unclear if he fell asleep while she was in the room or after she left. During STFU neither of Matthew's parents co-slept with him, representing an improvement from baseline.

Figure 6

Parental Responses Following Night Wakings across Baseline, Intervention and Follow-up Phases
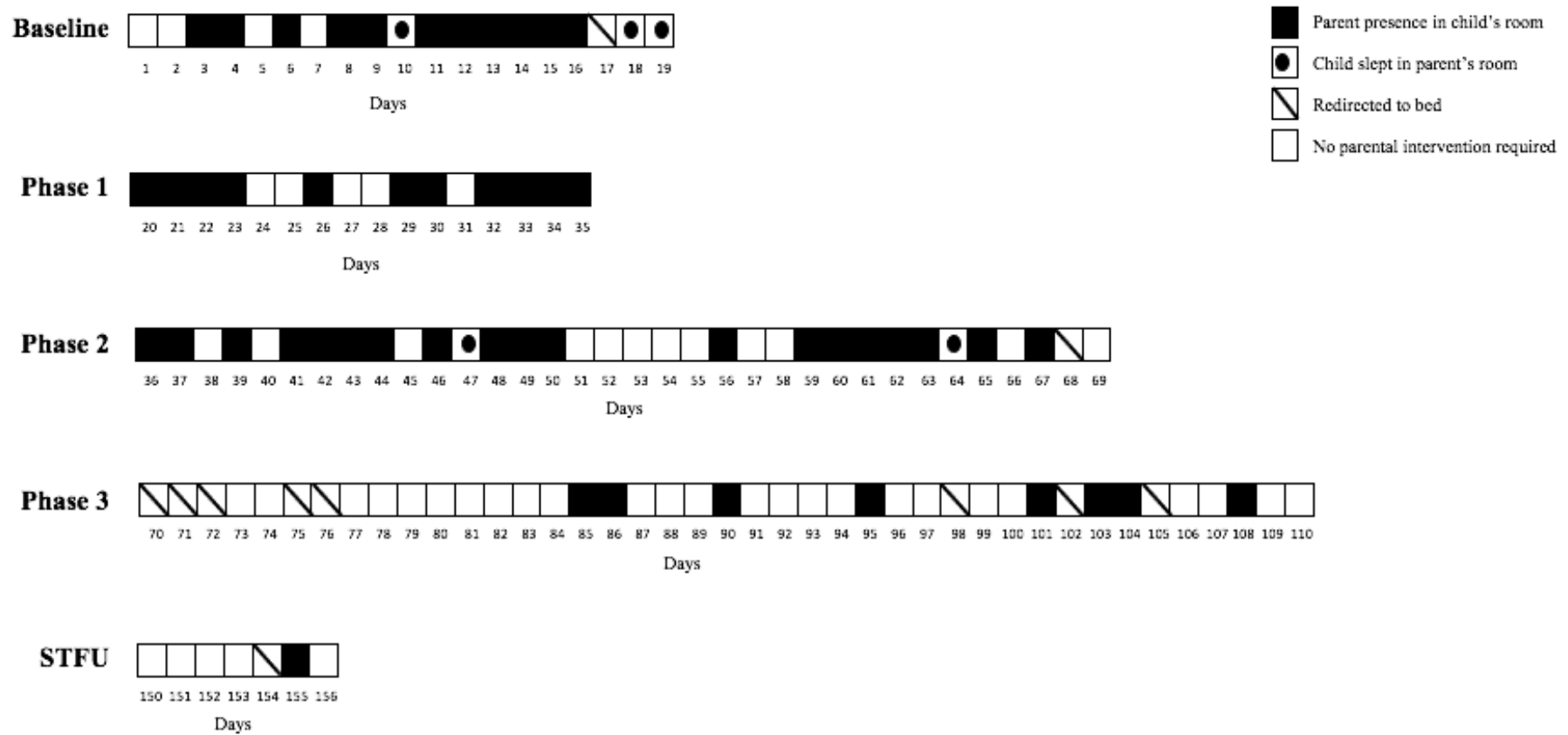
Early Morning Waking. The duration (mins) of time Matthew awoke before his scheduled/goal wake time across baseline, intervention and STFU phases are presented in Figure 7. During baseline, Matthew's wake time was 6.00am and EMW occurred on 11/19 mornings (58%), ranging from 3 – 35 minutes in duration. During Subphase 1 Matthew's wake time was adjusted to 5.45am (day 20), 6.00am (day 30), and then 6.15am (on day 36). This coincided with a decrease in EMW. Across all three intervention Subphases there were only two reported instances of EMW (2/91; 0.02%); one in Subphase 2 (day 40) and one in Subphase 3 (day 74). Day 40 (EMW of 57 mins) coincided with the family having begun their holiday the day before where Matthew spent four hours in the car, and on day 74 (EMW of 30 mins) he heard his father unwell in the next room. Throughout the remaining mornings during Subphases 1 – 3 Matthew often slept past his scheduled wake time and required his parents to wake him in the morning. During Subphase 1 his average wake time was 6.17am, in Subphase 2 it was 6.27am, and in Subphase 3 it was 6.35am, compared to an average wake time of 5.58am during baseline.

During STFU, no EMW occurred, and his average wake time was 6.46am. It is noteworthy that during STFU his parents maintained the consistent bedtime of 7.30pm (implemented at the end of Subphase 1). Thus the occurrence of no EMW during STFU represents a significant improvement as compared to baseline.

Figure 7

The Frequency and Duration of Early Morning Wakes across Baseline, Intervention and

Follow-up Phases
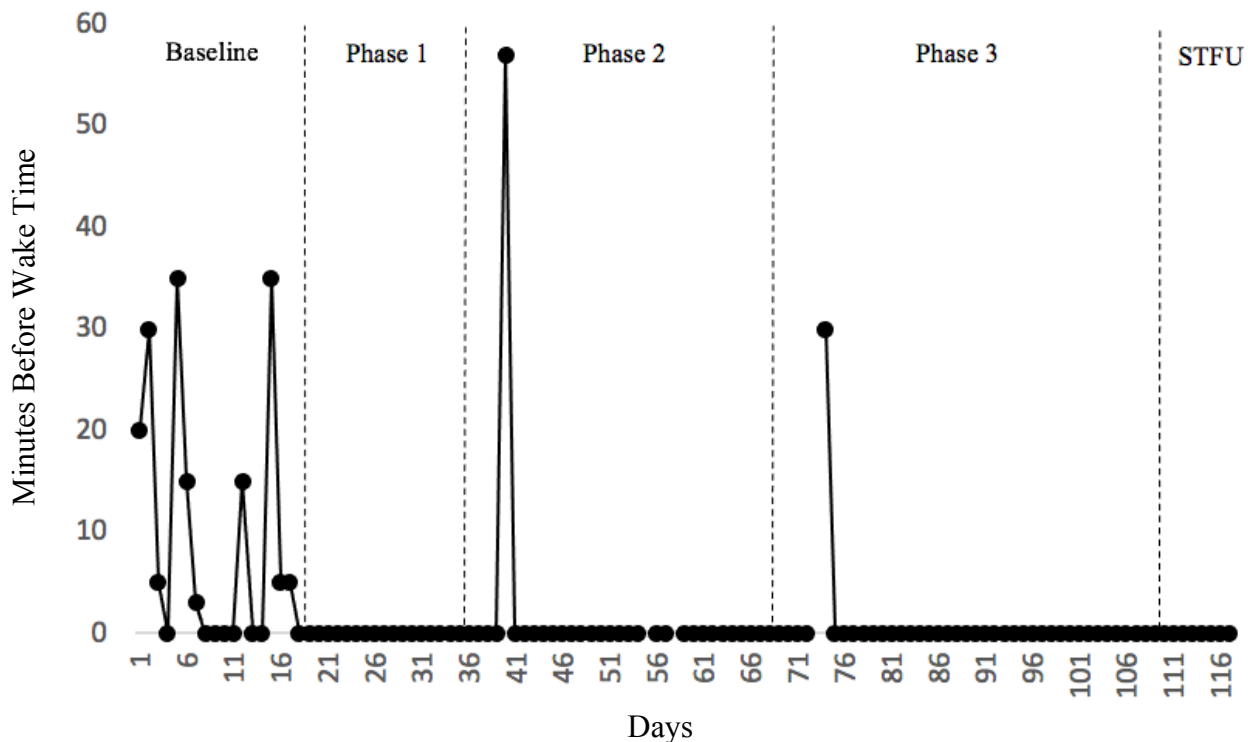
Psychometric Outcomes

Child Outcomes. The results for the CSHQ, the CBCL, and the PedsQL are presented in Tables 5 – 8 and are discussed below. These measures were all completed by Matthew's father.

The Child Sleep Habits Questionnaire. Results of the CSHQ are presented in Table 5. Higher scores on the CSHQ indicate a greater severity of sleep problems, and a score of > 41 indicates the presence of clinically significant sleep disturbance. At pre-intervention, Matthew's sleep problems were above the clinical cut off for sleep disturbance (score of 44). Following intervention, Matthew's total sleep difficulties score reduced to 37, representing a clinically significant improvement. There was also a reduction in the subscales of sleep onset delay (2 to 1), sleep duration (5 to 3), and NW (8 to 3). All other subscales remained stable,

except for daytime sleepiness which increased by one point. These scores indicate that the severity of Matthew's sleep difficulties, particularly NW, reduced following intervention.

Table 5

Comparison of Pre- and Post-Intervention Subscale and Total Scores on the CSHQ

Subscale	Pre-Intervention	Post-Intervention	Minimum Score
Bedtime Resistance	6	6	6
Sleep Onset Delay	2	1	1
Sleep Duration	5	3	3
Sleep Anxiety	4	4	4
Night Waking's	8	3	3
Parasomnias	7	7	7
Sleep Disordered Breathing	3	3	3
Daytime Sleepiness	9	10	8
Total Score	44	37	35

Note. Minimum Score = The minimum score possible on each subscale.

The Child Behaviour Checklist. Matthew's CBCL outcomes are presented in Table 6, with the raw scores presented for the eight subscales, and *T* scores presented for the Internalising, Externalising and Total problems scales. All pre- and post-intervention scores fell within a normal (i.e., non-clinical) range. Pre- to post-intervention comparison of Matthew's scores demonstrate improvements in the Internalising (*T* scores = 49 to 40), Externalising (*T* scores = 35 to 32) and Total Problems (*T* scores = 40 to 36) scales, indicating improvement. In particular, reductions occurred for the Somatic Complaints (3 to 2), Withdrawn (3 to 2), Sleep Problems (3 to 0), and the Aggressive Behaviour (2 to 1)

subscales. The Emotionally Reactive, Anxious/Depressed, Attention Problems and Other Problems subscale scores remained the same.

Table 6

Comparison of Pre- and Post-Intervention Syndrome Scale and Total Scores on the CBCL

Subscale	Pre-Intervention	Post-Intervention
Emotionally Reactive	1	1
Anxious/Depressed	0	0
Somatic Complaints	3	2
Withdrawn	3	2
Sleep Problems	3	0
Attention Problems	0	0
Aggressive Behaviour	2	1
Other Problems	3	3
Total Internalising	49 (N)	40 (N)
Total Externalising	35 (N)	32 (N)
Total Problems	40 (N)	36 (N)

Note. N = Normal range.

The Paediatric Quality of Life Inventory. The results of the PedsQL are presented in Table 7. The direction of beneficial change on the PedsQL is an increase in scores. A maximum score of 100 is possible for each subscale. At pre-intervention, Matthew was reported by his father to have relatively high HRQOL, with a total score of 79.3/100. Following intervention, this total score increased to 82.6/100, indicating a small overall improvement. Increases occurred on the Physical Functioning (90.6 to 100), Emotional Functioning (70 to 75), and Social Functioning (80 to 85) subscales, while the School

Functioning subscale decreased (70 to 60). At the time of post-intervention assessment, Matthew was reported to have missed more school owing to being unwell or going to the doctor/hospital compared to pre-intervention. His father noted that Matthew was absent for 30% of school days during the last school term.

Table 7

Comparison of Pre- and Post-Intervention Subscale and Total Scores on the PedsQL

Subscale	Pre-Intervention	Post-Intervention
Physical Functioning	90.6	100
Emotional functioning	70	75
Social Functioning	80	85
School Functioning	70	60
Total Score	79.3	82.6

Parent Outcomes. The results for the PSQI, DASS-21, and RQI are presented in Tables 8 – 10, and are discussed below.

The Pittsburgh Sleep Quality Index. The PSQI scores for Matthew's father are presented in Table 8. Higher scores reflect greater sleep impairment over the past month, and scores > 5 indicate poor sleep quality. At pre-intervention, the Total score (6) fell above the clinical cut-off and did not change at post-intervention. Post-intervention reductions were noted on the Subjective Sleep Quality (2 to 1), Sleep Duration (1 to 0), and the Sleep Efficiency (1 to 0) subscales, with scores remaining the same on the Sleep Latency, Sleep Disturbance, and Daytime Dysfunction subscales. The Use of Medication subscale increased (0 to 3); while Matthew's father noted that he had sustained a serious injury since the time of pre-intervention assessment, resulting in an increase in pain and medication use. Of note,

Matthew's father's TST increased (pre-intervention = 6.5 – 7 hours, post-intervention = 8 hours) demonstrating improvement. Overall, while the total score remained unchanged, results of the subscales were mixed, with no change seen in some subscales (3/7), improvement seen in some (3/7), and a decrease seen in one (1/7 = Use of Medication).

Table 8

Comparison of Pre- and Post-Intervention Subscale and Total Scores on the PSQI

Subscale	Pre-Intervention	Post-Intervention
Subjective Sleep Quality	2	1
Sleep Latency	1	1
Sleep Duration	1	0
Sleep Efficiency	1	0
Sleep Disturbance	1	1
Use of Medication	0	3
Daytime Dysfunction	0	0
Total Score	6	6

The Depression-Anxiety-Stress Scales. Results of the DASS-21 for Matthew's father are presented in Table 9. Higher scores indicate increased risk for emotional disturbance. Matthew's father reported no symptoms of depression, anxiety, or stress at pre- nor post-intervention (i.e., all scores of 0 fell within the normal range). Therefore, there were no changes in the subscale or total scores following intervention.

Table 9

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

Subscale	Pre-Intervention	Post-Intervention

Depression	0	0
Anxiety	0	0
Stress	0	0
Total Score	0	0
Sleep Efficiency	1	0
Sleep Disturbance	1	1
Use of Medication	0	3
Daytime Dysfunction	0	0
Total Score	6	6

The Relationship Quality Index. Results of the RQI for both parents are presented in Table 10. Higher scores represent greater relationship satisfaction, with the highest possible score being 45. Matthew's parents both reported a high degree of relationship satisfaction pre-intervention (father 38/45, mother 36/45), with Matthew's father reporting slightly higher satisfaction. At post-intervention, Matthew's father's score increased (40/45) and his mother's score decreased (34/45), indicating a small improvement and a small decrease in relationship satisfaction for Matthew's father and mother, respectively.

Table 10

Comparison of Pre- and Post-Intervention Total Scores for the RQI

	Father		Mother	
	Pre-Intervention	Post-Intervention	Pre-Intervention	Post-Intervention
Global Score	38	40	36	34

Treatment Acceptability. The outcomes of the post-treatment interview and the TARF-R are discussed below, with the TARF-R results presented in Table 11.

Post-Intervention Interview. Matthew's father described the intervention process as "ultimately a great process" and stated that it "made a massive impact to (their) household". This included that he felt that not only had Matthew's sleep improved, but also his own sleep and that of Matthew's mother. He reported that before the study they may have only slept through one night per week and both felt very tired throughout the day. Since the intervention, Matthew's sleep (and that of his parents') had only been disturbed 'within reason' and as parents they were both "less tired and grumpy".

Regarding the specific components of intervention, Matthew's father shared that the sleep/wake schedule changes were difficult to manage, reporting that although they understood the reasoning behind increasing sleep pressure, implementing the later bedtime was challenging to begin with because it meant shorter evening time together (mother and father). He also acknowledged that the presence of stressors external to the study (e.g., Matthew recently beginning school and increased risk of sickness, on-going concerns regarding COVID) had increased Matthew's levels of fatigue and as a result, it felt counter-intuitive to restrict his sleep. Matthew's father liked using the reward system which he thought resonated with Matthew. He also reported that while the rewards seemed important to Matthew's motivation during the initial stages of intervention, they were no longer required at the end. He felt that Matthew was proud of himself when he slept independently through the night and that this gave Matthew a sense of achievement. He also reported that the sleep diaries were easy to follow.

In terms of how the intervention process could be improved, Matthew's father believed there could have been a more holistic approach which better considered broader factors of their lives, as they initially found the intervention process overwhelming in the context of other child and family needs (e.g., Matthew's daily 90 mins of CF treatment and transitioning to school). For example, the recommended changes to Matthew's sleep/wake

schedule did not consider Matthew's exhaustion at the end of a school day. He believed that

if the clinician had been able to witness Matthew's treatment regimens and fatigue, they may

have better understood the wider context and impact of the recommended strategies. He did

however appreciate the flexibility of the clinician to meet the family's preferences regarding

adjusting the proposed sleep scheduling.

Overall, Matthew's father indicated that he felt positively about the intervention

process and was appreciative of the support from the clinician and the research team. He

noted that the value he places on research motivated the family to participate and contribute

to the study, and they were glad they persevered.

The Treatment Acceptability Rating Form – Revised. Table 11 displays the results of

the TARF-R for Matthew's father. Higher scores on the TARF-R illustrate better overall

treatment acceptability. Overall, the results demonstrate a good level of treatment

satisfaction, with a global score of 94/119. Results suggest that Matthew's father found the

treatment reasonable (19/21), that he was willing to carry out the intervention (17/21), that he

had a good understanding of the intervention process (6/7), and that he found the intervention

effective (21/21). Matthew's father also reported that the intervention did not result in

significant side effects (18/21), however, he did report that there was some cost involved with

the treatment (10/14). He also reported that the treatment was somewhat disruptive (9/21).

Table 11

Post-Intervention Subscale and Total Scores on the TARF-R

Subscale	Score	Maximum Score
Reasonableness	19	21
Willingness	17	21
Cost	10	14
Side-Effects	18	21

Effectiveness	21	21
Disruption/Time	9	21
Problem Severity*	6	14
Understanding of Treatment*	6	7
Total Acceptability Score	94	119

Note. * = not included in Total Acceptability Score

Discussion

The purpose of this case study was to investigate the effectiveness of a function-based behavioural sleep intervention for 5-year-old child with CF (Matthew), and the short-term maintenance of treatment effects. This study also investigated whether treatment would result in secondary benefits for Matthew and/or his parents, including his daytime emotions and behaviour and HRQOL, and parental sleep quality, wellbeing, and relationship satisfaction. Finally, this study also examined the social validity of the selected sleep intervention.

The primary sleep concerns for Matthew's parents were NW and reactive co-sleeping. Prior to intervention, Matthew woke up to four times per night, and was dependent on parental presence to re-initiate sleep following these NWs. His EMW were also considered to be a problem as he often woke earlier than his parents liked (before 5.30am). Although curtain calls and SOL were not parent-identified problems, these variables were also monitored as part of his overall sleep health. FBA revealed that his NW were positively reinforced by the presence of his parents in response in NW In addition, it was possible Matthew was experiencing a lack of physiological sleep pressure, resulting in fragmented sleep and his EMW. The primary goal of intervention was to enable Matthew to sleep through the night independently without requiring parental presence. The function-based intervention was introduced in stages according to the principles of minimal sufficiency,

whereby the hypothesised antecedent factors contributing to his sleep problems were targeted first. Intervention Subphase 1 involved sleep/wake rescheduling, Subphase 2 involved the implementation of a personalised social story and the use of reinforcement, and Subphase 3 involved limiting parent attention in response to NWs.

The Effectiveness of the Behavioural Sleep Intervention

Results of this case study show that the combination of behavioural sleep intervention strategies were effective in reducing the frequency and duration of Matthew's NWs, as well as his reliance on parental presence following NW. Matthew's NW steadily decreased throughout intervention, for example at baseline, NW occurred on 15/19 nights (79%), in Subphase 1 on 11/16 nights (69%), in Subphase 2 on 22/34 nights (65%), in Subphase 3 on 16/41 nights (39%) and in STFU on 2/7 nights (29%). The average duration of these NW also decreased (baseline = 18 mins, Subphase 1, 2 and 3 = all 9 mins, STFU = 10 mins). At baseline Matthew would require his parents to be present for him to reinitiate sleep following a NW, however from Subphase 3, Matthew was typically able to return to sleep independently after a NW. Despite NW continuing to occur intermittently throughout the intervention, his parents were satisfied that they occurred at reduced levels compared to baseline, and importantly, from Subphase 3, that he was able to resettle independently if he woke. This meant that his parents' sleep was also less disrupted following intervention. Therefore Matthew's improvements in the frequency and duration of NW, as well the amount of parental presence he required following NW represent a significant improvement from baseline, and these improvements were all maintained at STFU.

Improvement was also observed in the frequency of EMWs immediately following the implementation of Subphase 1. During baseline, Matthew woke before his scheduled wake time on 52% of mornings. However, following intervention EMW significantly reduced, with only two EMW occurring over the course of 91 days of intervention and seven

days of STFU. This again represents a significant improvement which was maintained at STFU.

Finally, during intervention and STFU, no treatment effects were observed for curtain calls nor SOL. Both SOL and curtain calls were non-problematic prior to intervention, and neither were specifically targeted by intervention. Thus it was not expected that an intervention effect would occur for these variables.

Improvement in Matthew's sleep was also reflected in his CSHQ scores. At baseline Matthew's score (44) was above the clinical cut-off (41). Following intervention, this score reduced to 37. Item analysis showed that the NW subscale decreased from 8/9 at baseline to 3/9 (the lowest possible score) post-intervention. This reflects Matthew's father's perception that NW were no longer problematic, which is an important real-life measure of the significance of Matthew's improvement.

Additionally, the intervention resulted in small improvements in Matthew's daytime emotions and behaviour (measured by the CBCL), as his total Internalising, Externalising and Problems scores increased. Additionally, Matthew's total HRQOL improved (measured by the PedsQL), with all subscales increasing apart from the subscale of School Functioning, as Matthew's father noted he had missed a significant amount of school recently due to being unwell or needing to go to the Doctor. There was also some improvement in Matthew's father's sleep (measured by the PSQI), with the scales of Subjective Sleep Quality, Sleep Duration and Sleep Efficiency improving, while all other scales remained unchanged, apart from the Use of Medication subscale due to his recent injury. There were no significant changes to his parents relationship quality (measured by the RQI) or his father's mental wellbeing (measured by the DASS-21). Finally, the intervention was described by Matthew's father as acceptable, effective, and socially meaningful. Overall, results of this study provide preliminary evidence of the effectiveness and social validity of a behavioural sleep

intervention for a child with CF, and suggest that improving sleep may be associated with a range of secondary benefits.

These findings are in line with previous research discussed in Chapter Two (Kellerman, 1979; Bartlet & Beaumont, 1998; Wiggs & Stores, 1998; Willgerodt et al., 2014; Palermo et al., 2016, 2017; Zhou et al., 2017; Zupanec et al., 2017; Law et al., 2018; Tumakaka et al., 2019; Sonney et al., 2020; Tsai et al., 2020; Zhou & Recklitis, 2020), which showed that sleep problems in children with various CHC, including respiratory conditions (Bartlet & Beaumont, 1998; Willgerodt et al., 2014; Palermo et al., 2016, 2017; Sonney et al., 2020) also improved following psychosocial sleep interventions. Although many of the studies discussed in Chapter Two included older children, six studies (Kellerman, 1979; Bartlet & Beaumont, 1998; Zupanec et al., 2017; Sonney et al., 2020; Tumakaka et al., 2019; Tsai et al., 2020) included children of a similar age to Matthew. For example, Tsai et al. (2020) demonstrated improved sleep quality and quantity in children with epilepsy of a similar age ($N = 100$, M age= 3.87 years) using similar behavioural strategies of psychoeducation and sleep/wake scheduling. Although Tsai et al. (2020) did not focus on NW and instead aimed to improve SE, nighttime TST and the overall daily sleep duration (for children that also required naps), a similar approach was used where parents were educated on strategies to promote sleep, including implementing appropriate sleep habits and sleep schedules. Tsai et al. (2020) found that the intervention resulted in improved sleep quality and quantity within their intervention group, therefore like the current case study, demonstrating how behavioural sleep interventions can be effective in improving sleep within young children with CHC. This case study therefore extends the results of previous studies to provide preliminary support for the effectiveness of psychosocial sleep interventions in a child with CF.

The introduction of each intervention component within the current study appears to have differentially affected Matthew's sleep. Following the implementation of sleep/wake scheduling in Subphase 1, the frequency of EMW immediately reduced, and the duration of NW also significantly decreased. This effect was largely maintained throughout intervention and STFU. Importantly, Matthew's parents were consistent with the scheduled bedtime throughout the intervention which meant Matthew's TST also remained stable from baseline to STFU. Thus, the changes in his sleep/wake schedule did not impair the amount of sleep he obtained each night. Previous research has found that delaying bedtimes in healthy children and children with developmental disabilities can reduce the frequency and duration of NW, however, doing so has often resulted in a reduction in TST (Piazza et al., 1998; Sadeh et al., 2003; Christodulu & Durand, 2004; Durand & Christodulu, 2004). Furthermore, no previous research could be identified which focused solely on delaying bedtimes to target EMW, despite the theoretical underpinnings of this strategy being well accepted. Thus, the current case study adds to the current literature on sleep/wake scheduling by providing an example of an effective method of reducing EMW in a child with CF.

The implementation of the personalised social story and reinforcement in Subphase 2 also saw a continuation of reductions in the frequency of NW from Subphase 1. Although the improvements from Subphase 1 to Subphase 2 were not significant, it is possible that there may be some cumulative benefit to the use of these strategies. It is not possible, however, to isolate treatment effects to determine whether the use of these strategies in isolation (i.e., without sleep/wake rescheduling) would have been effective. Previous studies have utilised social stories within behavioural sleep interventions for children with developmental disabilities, finding positive outcomes (Moore, 2004; McLay et al., 2017, 2019; Van Deurs et al., 2019; Hunter et al., 2021). This includes a study in which social stories were the sole intervention strategy (Moore, 2004), as well as when used in combination with other

behavioural sleep intervention strategies (McLay et al., 2017, 2019; Van Deurs et al., 2019; Hunter et al., 2021). One study by Burke et al. (2004) investigated the use of social stories in isolation (i.e., no other intervention was included other than rewards) among four neurotypical children (aged 2 – 7 years) to address bedtime resistance and NW. The stories detailed parental expectations for appropriate bedtime behaviours and the reward the child would receive for meeting those expectations. Burke et al. (2004) found that the social story produced a reduction in disruptive bedtime behaviours and NW, which were maintained at a 3-month follow-up. Thus demonstrating the effectiveness of these techniques as an intervention component and providing support for their use in the current study.

Previous research has also suggested that the use of positive reinforcement (e.g., through tangible rewards) as a component used in combination with other psychosocial sleep intervention strategies is useful for improving children's sleep (Meltzer, 2010). However, as in social stories, it is difficult to establish whether the use of reinforcement alone would have been effective for Matthew. Studies have found that as a component, used in combination with other psychosocial sleep intervention strategies, reinforcement is useful for improving children's sleep. For example, Ollendick et al. (1991) investigated the use of CBT for decreasing bedtime fears in two children (8 and 10 years) experiencing separation anxiety. Their intervention used a multiple-baseline-across subjects design, including two treatment phases: self-control training alone, and self-control training with contingent reinforcement. Like the present case study, the reinforcement component included rewards for nights in which the children remained in bed alone for the entire night. Ollendick et al. (1991) found that although the self-control-only phase resulted in a slight reduction in the children's anxiety, the introduction of reinforcement resulted in a marked increase in improvement. Although it is not possible to determine from the present case study whether the use of reinforcement alone would have been effective for Matthew, the study by Ollendick et al.

(1991) demonstrates that reinforcement may be an important component to enhance other intervention strategies.

Finally, in Subphase 3, there was a notable reduction in the frequency of NW compared to previous phases following the removal of parental presence and restricting parent attention to NW-behaviour. By the end of Subphase 3 Matthew was no longer constantly dependent on parental presence to return to sleep following NW. The removal of parental presence (i.e., extinction) coupled with redirecting him to bed likely contributed to Matthew learning to resettle independently over time. Thus this strategy can be seen as an effective intervention component which contributed to a significant improvement in Matthew's NW and the impact they had on his parents.

Matthew's co-sleeping could be understood in terms of operant conditioning, where his behaviour of going to his parents' room when he woke was reinforced by parent attention (co-sleeping). Although co-sleeping can mitigate sleep problems in the short term (Keller & Goldberg, 2004; Goldberg & Keller, 2007; Ramos et al., 2007; McLay et al., 2019), it meant that he developed sleep onset associations where he was dependent on his parents' presence to resume sleep. The skill to settle to sleep independently is important, as children who do not possess this skill are likely to have ongoing difficulties with sleep (Meltzer, 2010). Thus it was important to rectify Matthew's difficulty with self-settling in order to not only reduce the impact it had on his sleep, but also his parents' sleep.

Previous studies have demonstrated the efficacy of extinction and reinforcement methods including the 'excuse me' drill to reduce co-sleeping and NW in children (Kuhn, 2011). The procedures used in this case study were similar to the 'excuse me' drill which combines extinction (i.e., minimising parental attention for sleep-competing behaviours) and reinforcement (e.g., praise for sleep-conducive behaviours) to teach children to initiate sleep independently (Kuhn, 2011). Importantly, as was the case with Matthew, the excuse me drill

is only effective when the child's sleep difficulties are maintained by the reinforcing properties of parental presence or attention (Kuhn, 2011). Kuhn et al. (2019) investigated the effectiveness of the excuse me drill in promoting independent sleep onset in four typically developing children (aged 2 – 7 years). Following the implementation of the excuse me drill, all four children rapidly learnt to initiate sleep independently (Kuhn et al., 2019). The findings of the present case study therefore provide some additional support for the effectiveness of the principles of the excuse me drill as a strategy for teaching children to fall asleep independently.

Overall, the findings of this case study are consistent with previous research showing that behavioural interventions can effectively reduce sleep problems in children, including typically developing children and those with developmental disabilities, and CHC (Kellerman, 1979; Ollendick et al., 1991; Bartlet & Beaumont, 1998; Wiggs & Stores, 1998; Burke et al., 2004; Sadeh et al., 2003; Durand & Christodulu, 2004; Moore, 2004; Willgerodt et al., 2014; Palermo et al., 2016, 2017; McLay et al., 2017; Zhou et al., 2017; Zupanec et al., 2017; Law et al., 2018; Kuhn et al., 2019; McLay et al., 2019; Tumakaka et al., 2019; Van Deurs et al., 2019; Sonney et al., 2020; Tsai et al., 2020; Zhou & Recklitis, 2020; Hunter et al., 2021). In particular, Lunsford-Avery et al. (2021) conducted a systematic review to assess the efficacy of behavioural sleep interventions for sleep disturbances in school-aged children (aged 6 – 18 years) who were either typically developing or had neurodevelopmental difficulties. They reported that the effectiveness of interventions varied according to the mode of delivery, intervention content, and target population. Specifically, these interventions were reported to be more likely to be effective if delivered by an individual, rather than solely through printed materials. The importance of individualised interventions tailored to the child/family were also discussed by Lunsford-Avery et al. (2021), as individualised interventions were found to be effective immediately following intervention

and in the long-term (i.e., 3 – 6 months post-treatment), including in reducing NW and the likelihood of the child sleeping in their own bed. Likewise, the study by Bartlet and Beaumont (1998) which was discussed in Chapter Two found that individualised behavioural strategies, including extinction and reinforcement strategies were effective in improving settling and NW among children with developmental disabilities and children with CHC (including chronic upper-respiratory tract infections, eczema, deafness, asthma, epilepsy, and coeliac disease).

Finally, the study by Sonney et al. (2020) which was also discussed in Chapter Two, piloted a study to examine the effectiveness of an online CBT-based sleep intervention they developed, Sleep Intervention for Kids and Parents (SKIP) in 25 children (ages 6-11 years) with asthma. Their study was similar to the current case study, in the sense that the intervention children received was recommended based off their unique sleep difficulties, as opposed to a generic intervention aimed at broad sleep difficulties. Sonney et al. (2020) found that their intervention resulted in various improvements in the children's sleep (e.g., improved SE and reduced WASO) and that treatment effects were maintained at follow-up. These findings are in line with those of the current case study as they also suggest that individualised psychosocial sleep interventions can effectively reduce sleep difficulties in young children with chronic respiratory conditions. Importantly, the study by Sonney et al. (2020) also illustrates how sleep difficulties in children with respiratory conditions are not always due to unmodifiable consequences of their illness.

When interpreting the results of this case study, it is important to consider various factors that may have continued to influence Matthew's sleep throughout intervention. As discussed earlier, a number of biopsychosocial factors can contribute toward sleep problems in children with CF and may in turn, influence response to behavioural sleep interventions (Canter et al., 2021). For example, for Matthew there were various instances during

intervention in which he was unwell and required the administration of pain relief. In healthy children, periods of being unwell often coincide with sleep difficulties (Mitra et al., 2011). As such, it is likely for Matthew that regardless of the overall improvements seen from the behavioural sleep intervention, NW may still continue to some degree for reasons related to his condition. Improvements in sleep among children with CF may therefore not always appear linear, as illness related factors will likely contribute to problem's reemerging spontaneously. Because of these factors, intervention goals for children with CF and sleep problems need to be realistic, and like in Matthew's case, it may be more important to reduce NW rather than aim to completely eliminate them, especially during periods where health is more affected. It is also important to note that despite Matthew having periods of increased sickness throughout the course of intervention, his sleep problems did not revert to baseline levels. This suggests that modifiable factors (e.g., parent attention) were contributing to and maintaining his sleep difficulties aside from illness-related factors. It is therefore important that consideration is given to health needs and illness-related factors when considering treatment goals and how effective an intervention will be for children with CF (e.g., NW may be reduced rather than eliminated). Additionally, careful consideration also needs to be given to the role of medical factors (e.g., CF treatments) during assessment and intervention processes as these also may impact the child and family's abilities to carry out sleep intervention strategies.

Secondary Outcomes of Intervention for Matthew

The CBCL and PedsQL were used to investigate whether the selected behavioural sleep intervention would improve Matthew's daytime emotions, behaviour, and HRQOL. Previous research has shown that sleep problems are associated with increased risk of somatic complaints, aggression and social problems in children (Simola et al., 2014), and that poor sleep is associated with poor HRQOL and lowered emotional and behavioural functioning in

children with CF (Dancey et al., 2002; Bouka et al., 2012; Forte et al., 2015; Íscar-Urrutia et al., 2018; Shakkottai et al., 2018; Tomaszeck et al., 2018; Vandeleur et al., 2018). Furthermore, research has shown that the use of behavioural sleep interventions in typically developing children and children with developmental disabilities has led to improvements in sleep and associated improvements in daytime behaviours and psychological functioning (Hiscock et al., 2014; Blake et al., 2017; Hunter et al., 2020). Following intervention for Matthew, there were small increases in Internalising and Externalising symptoms as well as Total Problems scores on the CBCL, which included increases on the sleep subscale. Additionally, there was a small improvement in Matthew's total score on the PedsQL. Small changes might be expected given that at pre-intervention the CBCL scores fell within a normal range and Matthew was reported to have high HRQOL (i.e., a ceiling effect). Nevertheless, these changes suggest that behavioural sleep interventions may also produce untargeted benefits to daytime functioning.

These findings are supportive of the results of studies discussed in Chapter Two (Willgerodt et al., 2014; Palermo et al., 2017; Zhou & Recklitis 2020), where improvements in psychological wellbeing and daytime behaviours were found following psychosocial sleep interventions. However it is important to note that unlike the present study, for two of these studies (Palermo et al., 2017; Zhou & Recklitis) cognitive therapy was included, thus wellbeing symptoms were not necessarily considered untargeted/collateral outcomes. Nevertheless, Palermo et al. (2017) found that in adolescents with sleep problems and co-occurring medical and mental health conditions, a CBT-I intervention resulted in improvements in sleep, as well as reductions in anxiety (as measured with the Paediatric Anxiety Short Form), depression (as measured with the Paediatric Depressive Symptoms Short Form), and HRQOL (as measured with the PedsQL). Additionally, Willgerodt et al. (2014) discovered that their MBI intervention for children with sleep problems (including

children with asthma) resulted in improved sleep and improvements in parent-reported child problem behaviour scores (as measured with the Eyberg Child Behavior Inventory [ECBI]). Finally, Zhou and Recklitis (2020) found that their online sleep intervention programme resulted in improvements in sleep and HRQOL (as measured on the PrdsQL) in adolescent and young adult cancer survivors. It is however important to acknowledge that the mechanisms underlying the improvements in Matthew's daytime emotions, behaviour and HRQOL cannot be determined, as a multitude of other factors may have contributed to these improvements (i.e., potential improvements in physical health, family factors). Caution notwithstanding, the current case study adds to previous research by demonstrating that untargeted improvements in daytime functioning and wellbeing may occur for children with CF following a behavioural sleep intervention. .

Secondary Outcomes of Intervention for Matthew's Parents

The PSQI, DASS-21, and RQI were used to assess change in Matthew's father's sleep, wellbeing, and in parental relationship satisfaction, respectively. Matthew's father's overall PSQI score remained unchanged following intervention, however when looking at specific subscales of the PSQI, there were improvements in some variables (i.e., Subjective Sleep Quality, Sleep Duration and Sleep Efficiency), and when taking into consideration his explained increase in medication use, his overall sleep improved. There were however no changes in Matthew's father's reports of symptoms of depression, anxiety and stress (as measure on the DASS-21) from pre- to post-intervention. Additionally, prior to intervention, Mathew's parents' scores on the RQI reflected that they were satisfied in their relationship, and there were only minor changes to these scores at post-intervention.

The most significant changes in Matthew's father's wellbeing post-intervention were in regard to the improvements seen in some subscales of sleep as measured by the PSQI, and importantly the increases in his reported TST (from 6.5 – 7 hours pre-intervention, to 8 hours

post-intervention). One plausible explanation for the improvements in Matthew's father's sleep following intervention is that improvements in Matthew's sleep (e.g., reduction of NW, EMW, and ability to settle independently following NW) resulted in improvements in his own sleep quality and/or sleep quantity. The fact that Mathew's father's sleep was impaired at baseline is in line with extant research that consistently shows that parents' sleep is negatively impacted when attending to a child's NW (National Sleep Foundation, 2004, Meltzer & Montgomery-Downs, 2011; Byars et al., 2020). For example, in a U.S. national survey of children's sleep habits, behaviours and difficulties, more than 50% of surveyed parents reported losing an average of 30 minutes sleep each night due to their child's NW (National Sleep Foundation, 2004; Meltzer & Montgomery-Downs, 2011). This therefore suggests that it is also important to assess parents' sleep quality when assessing children's sleep problems.

The improvements in Matthew's father's sleep following intervention are also consistent with the findings of other behavioural sleep intervention studies. For example, the study by Sonney et al. (2020) which was discussed in Chapter Two, found that their online sleep intervention not only improved the sleep of their 25 participants (children with asthma), but also their parent's sleep. Specifically, following intervention, there were improvements in WASO, SE and the variability in bedtimes for parents (Sonney et al., 2020). Further, another study discussed in Chapter Two by Wiggs and Stores (1998) found that a tailored behavioural sleep intervention improved the sleep of 30 children (including children with epilepsy) with challenging daytime behaviour, and also resulted in secondary benefits for their mother's sleep. Furthermore, significant associations were found between the TST of children and their mothers' sleep (Wiggs & Stores, 1998). Thus, these studies illustrate the interconnected relationship between the sleep of children with CHC and their parents, which was also demonstrated within the current case study.

Although extant research has demonstrated that depression and anxiety are more prevalent in parents of children with CF as compared to parents of typically developing children (Glasscoe et al., 2007; Wong & Heriot, 2008; Yilmaz et al., 2008; Driscoll et al., 2009; Smith et al., 2010; Besier et al., 2011; Quittner et al., 2014; Duff, 2015; Barker & Quittner, 2016; Cronly et al., 2019; Lord et al., 2022), Matthew's father did not indicate symptoms of anxiety or depression were of concern to him. This finding is important as previous research has demonstrated associations between poor sleep in children with CF and poorer parental wellbeing (Sivertsen et al., 2009; McQuillan et al., 2019; Byars et al., 2020), which does not appear to have occurred within the current case study. Again, like Matthew's psychological functioning this lack of change may also relate to a floor effect where Matthew's father's psychological functioning pre-intervention was already within the normal range, leaving no room for improvements. Therefore, no conclusions can be drawn regarding the impact of intervention on Matthew's father's psychological functioning.

While there was little change in Matthew's father's psychological functioning or his parent's reported relationship quality, it is nonetheless important to acknowledge that many variables not necessarily captured within the current case study can affect parental wellbeing. For example, the health of parents and the health of their child/children, as well as employment status, financial stress and the number of children in a household can contribute to parent's reported levels of psychological distress and/or their level of relationship satisfaction.

It is also important to consider the role that parent wellbeing may play in relation to children's sleep problems. For example, research shows that parental stress is associated with a higher likelihood of parents responding to their children's sleep difficulties in ways that are not conducive to promoting quality sleep (Johnson & McMahon, 2008; Coto et al., 2018; Tyler et al., 2019). For example, Johnson and McMahon (2008) investigated the associations

between parental hardiness, sleep-related cognitions and bedtime interactions in 110 parents of preschool children (*M* age = 3.81 years) through a survey. They found that parental hardiness, which refers to parent's ability to manage their children's challenging behaviors effectively by transforming stressful situations into opportunities for learning and personal development (Maddi, 2002), predicted children's sleep problems, even after controlling for temperament (Johnson & McMahon, 2008). Parents' attributions and behaviour (in terms of how they perceive and respond to children's sleep behaviour) are also major determinants of the success of behavioural sleep interventions, as parents are often responsible for enforcing intervention strategies, especially within young children (McLay et al., 2020). It is therefore important to consider parenting factors, such as levels of stress, as these factors may influence parents' ability to change their responses to children's sleep problems and adhere to a behavioural programme.

Social Validity of the Behavioural Sleep Intervention

The post-intervention interview and the TARF-R were used to investigate the acceptability of the selected behavioural sleep intervention for Matthew's father. Results suggest that Matthew's father found the intervention acceptable, effective, understandable, and socially meaningful. This finding is consistent with previous research which shows that behavioural sleep interventions are considered reasonable, understandable, effective, and acceptable to parents of children with developmental disabilities (McLay et.al., 2019; McLay et.al., 2020; Van Deurs et.al., 2021), and parents of children with CHC (Law et al., 2018; Sonney et al., 2020; Willgerodt et al., 2014). Specifically, as discussed in Chapter Two, Law et al. (2018) reported that adolescents and parents in their study found their CBT-I intervention to be acceptable (moderate score on the TEI-SF), Sonney et al. (2020) reported that 92% of parents in their study were satisfied with their online sleep intervention, and Willgerodt et al. (2014) reported that parents in their study found the clinical support of their

MBI intervention to be acceptable and not burdensome. Thus, as reported by Matthew's

father and supported by previous research, although behavioural sleep interventions may

require a significant commitment from families, they are generally perceived to be worthy of

the time and effort. Importantly, Matthew's father noted the significant positive impact the

intervention had for his family, including how improvements in their own sleep as parents

had improved their daytime functioning, thus demonstrating further important secondary

impacts of the intervention.

One specific challenge noted by Matthew's father was regarding the difficulty they

experienced adjusting Matthew's bedtime, as it shortened evening time for them as parents.

Furthermore, Matthew's father was concerned about Matthew's level of fatigue in the context

of settling into school. Matthew's father's reports of finding the intervention difficult to

manage at times are line with the findings of Bartlet and Beaumont (1998). In the study by

Bartlet and Beaumont's (1998) discussed in Chapter Two, they used individualised

behavioural sleep strategies to decrease NW and settling difficulties in children who were

chronically ill or had developmental disabilities. They found that some families (7/61)

reported the intervention to be difficult to manage, although many of the families did in fact

report the intervention to be useful (27/61). This demonstrates how behavioural sleep

interventions may not be uniformly acceptable to all parents of children with CHC.

Nonetheless, despite the challenges of the current intervention, the overall feedback from

Matthew's father was positive.

One recommendation from Matthew's father was that a more holistic approach that took

into consideration other child and family factors could have improved the intervention. This

recommendation is in line with previous research that has suggested due to the unique

clinical characteristics of CF, sleep interventions need to take into consideration various

factors including treatment burden (Canter et al., 2021). Although clinicians should aim to

reduce intervention and research burden (e.g., data collection) for all families, for children with CF this may be especially important as symptom management is a key priority. Thus, because of the need to put the child's immediate health before improvements in their sleep, it is possible that illness may impact a family's ability to adhere to behavioural sleep intervention processes. This has been illustrated in previous research. For example, in the aforementioned study by Bartlet and Beaumont (1998), illness, particularly respiratory infections, was reported to delay the intervention process. Likewise, in the study by Law et al. (2018), four children did not complete the intervention due to a major health event. Thankfully within the current case study, despite Matthew experiencing frequent periods of illness, he did not experience any severe illness exacerbations that could have impaired his family's ability to complete the intervention.

Importantly, in terms of treatment fidelity, Matthew's parents remained consistent with implementing the treatment components throughout the intervention process. Previous research has indicated that overall treatment effectiveness is linked to treatment adherence (Riedel & Lichstein, 2001; Vincent & Hameed, 2010), thus Matthew's father's high motivation to commit to the intervention programme, his understanding of the intervention, and perceived acceptability of the intervention, likely contributed to the improvements in Matthew's sleep.

Limitations

There are several limitations to the case study. First, it lacked objective sleep measures (e.g., actigraphy and/or videosomnography). As a result, it was not possible to triangulate subjective and objective data, nor to calculate Inter-rater Observer Agreement (IOA). The absence of IOA may therefore threaten the internal validity of findings (Didden et al., 2002; Ledford & Gast, 2009; Watkins & Pacheco, 2000). A reliance on sleep diaries also meant that it was not possible to objectively determine whether parents implemented the

intervention with fidelity. For example, it was not possible for the clinician to determine if parents deviated from or did not implement components of the intervention plan (including subphases) as was recommended. Additionally, previous research has found that objective measures of sleep and parent-report provide differing, but complementary, information regarding a child's sleep behaviour (Holley et al., 2010; Dayyat et al., 2011). For example, objective measures may provide information regarding the occurrence of NW which parents may be unaware of (e.g., if the child does not signal that they are awake), whereas parent-report may provide more details regarding specific behaviours during NW, especially if the child is out of the range of the camera. Thus, the case study data are limited to that which parents were aware of.

Sole reliance on parent-report means there is a possibility of bias in parent-reported data (Dayyat et al., 2011). This limitation also applies to how the psychometrics within the case study relied on parent-report, and therefore may have been vulnerable to response bias and inaccurate reporting (Weiskop et al., 2005; Loring et al., 2018). Furthermore, secondary results were limited to the focus of psychometric measures that were chosen for inclusion, which may have meant that secondary changes occurred that went unmeasured. For example, the case study did not include a measure of CF-specific symptoms, thus changes in Matthew's physical health were not able to be assessed.

Another limitation is that only a single person was included in the case study. As such, it is difficult to make inferences regarding the effectiveness of psychosocial sleep interventions for other children with CF, including children of different ages, ethnicity and levels of illness severity. For example, as Matthew was young and pancreatic sufficient, his potential illness burden may have been less than some other children with CF (Stephenson et al., 2017). It is important to include multiple participants within single-case research design research (e.g., a multiple baseline design) where possible, because replication of treatment

effects (e.g., across participants) is a critical component of determining internal validity (Kratochwill et al., 2013).

The intention of the current research was to include multiple children, however, recruitment difficulties meant this was not possible. This may reflect unique challenges associated with undertaking psychosocial sleep intervention research with children with CHC, including CF. According to the New Zealand CF Registry, there were only 247 children (aged 0-18 years) with CF living in New Zealand in 2017 (Cystic Fibrosis NZ, 2017), thus representing a small population for recruitment. In addition, in order to be eligible for inclusion, families not only had to be willing to participate, but children were required to be experiencing sleep difficulties during a period of illness stability. Families may have also chosen not to partake in the research due to the assumed burden, as there are already immense time-consuming illness demands for families living with CF (Sawicki et al., 2012; Hafen et al., 2013; Jennings et al., 2014).

Finally, considering the number of children impacted by CHC (including CF) worldwide, and that these numbers are only predicted to continue to increase (Perrin et al., 2007), the evidence base for the use of psychosocial sleep interventions for this population is very small. Research has demonstrated that these children experience high rates of sleep problems which have the potential to worsen their physical symptoms (Bryant et al., 2004; Banks & Dinges, 2007; Lewandowski et al., 2011; Short & Banks, 2013; Medic et al., 2022); therefore, it is important that further research is conducted in this area, because the current breadth of the literature does not fill the research gap.

Conclusion

The overall findings from the case study demonstrate that the selected behavioural sleep intervention was effective in improving Matthew's the frequency and duration of NW and EMW. Although NW did not completely resolve, the improvements following

intervention were considered socially significant to his parents. Importantly, there was a reduction in the frequency that Matthew's parents' co-slept with him following intervention, which was a socially meaningful improvement for the family. These improvements were also all maintained at STFU.

There were no changes in Matthew's SOL nor curtain calls following intervention, however this may be expected given that SOL was at reduced (non-clinical) levels during baseline, and curtain calls, which were considered non-problematic by parents, were not directly targeted. Additionally, small improvements were seen in Matthew's daytime emotions and behaviours, as well as his HRQOL. There were also some improvements in Matthew's father's sleep. No changes occurred for symptoms of depression, anxiety and stress, or for parental relationship satisfaction. Finally, the intervention was described as effective and acceptable by Matthew's father. Thus, the intervention resulted in a range of positive outcomes. These outcomes should however be considered in the context of various limitations, including the reliance on subjective measures (sleep diaries and psychometrics), and the inability to generalise findings due to only having one participant. The key findings of the case study, strengths of this research, clinical implications and directions for future research are discussed further in Chapter Four, in relation to the findings from Chapter Two.

Chapter Four: General Discussion

Children with CF are at an increased risk of experiencing sleep disturbance, which may in turn impact their overall wellbeing and ability to manage their illness demands (Shakkottai et al., 2018). To date, there has been little research exploring the effectiveness of psychosocial sleep interventions for children with CF. Therefore, it is important that further research is undertaken to inform our understanding of the effectiveness and appropriateness of psychosocial interventions to treat sleep problems in children with CF. The overarching aim of this thesis was to investigate the use of psychosocial (including behavioural) sleep interventions for children with CF, which was done through two studies. This chapter begins by summarising the systematic review (Chapter Two) and case study (Chapter Three). This chapter will then discuss the key findings of the review and case study in relation to the overall research aims of this thesis. The strengths and contributions of this research are then considered. Finally, this chapter discusses clinical implications of the overall findings, directions for future research, and provides an overall conclusion.

Summary of Chapters Two and Three including Key Findings

Chapter Two presented a systematic review, which was conducted to identify and evaluate the use of psychosocial sleep interventions to treat sleep problems in children with CHC. This review aimed to: (1) identify and appraise the literature examining the effectiveness, feasibility and social validity of psychosocial sleep interventions for children with CHC; (2) identify how these findings can inform the design of future research examining the effectiveness of psychosocial sleep interventions for children with CHC, including CF; and (3) identify how these findings can inform assessment and intervention processes used in clinical practice with children with CHC, including CF, and their parents/carers. The systematic search identified 13 studies that met inclusion criteria. These studies were then evaluated in terms of their key intervention components (e.g., participant

characteristics, methodologies, inclusion of follow-up), the outcomes of intervention, and the methodological rigor of the study, according to the Reichow et al. (2008) evaluative method. The reviewed studies utilised a variety of psychosocial sleep intervention techniques, including behavioural (e.g., stimulus control, extinction techniques, and reinforcement) and Cognitive Behavioural Therapy for Insomnia (CBT-I; e.g., cognitive restructuring, relaxation training) strategies. Intervention agents included parents, adolescents/adults or parent-adolescent dyads. Intervention content was delivered in a variety of ways including in-person and online, and interventions were implemented in the home-setting or clinic.

The results of this review revealed that a majority of the studies (12/13) reported at least some positive sleep effects across participants. This included reductions in Sleep Onset Latency (SOL), Wake After Sleep Onset (WASO), Night Wakings (NW), Early Morning Waking (EMW) and improvements in sleep hygiene and overall sleep quality. Thus, the findings were in line with the literature exploring similar interventions within typically developing children and children with neurodevelopmental disorders. Five studies also investigated secondary intervention effects, with all five reporting some aspect of secondary improvements, including in Quality of Life (QoL), daytime problem behaviours, symptoms of anxiety and depression, and headache frequency (for children with chronic migraine). The majority of treatment effects (sleep and secondary outcomes) were also reported to be maintained at follow-up. Furthermore, treatment satisfaction was generally reported to be moderate to high for the eight studies which reported this. Treatment fidelity was reported by four studies and was also generally reported as high in three of these studies. Although these findings are promising, only 4/13 studies met criteria to be classed as having 'strong' or 'adequate' methodological quality, while the remaining nine studies received a 'weak' quality rating according, to the Reichow et al. (2008) evaluative method.

Despite the need to address sleep problems in children with CHC, the review concluded that there is a lack of research investigating the effectiveness of psychosocial sleep interventions for children with CHC. The high number of reviewed studies lacking strong methodological rigor limits our certainty in the evidence available, as well as the ability to generalise findings. Further research of strong methodological quality is needed to replicate and extend treatment effects. Further research is also needed which investigates the effectiveness of psychosocial sleep interventions across a broader range of CHC, as one limitation of the research reviewed was that a limited range of CHC have been investigated. For example, no studies were identified that examined the effectiveness of psychosocial sleep interventions for children with CF.

The outcomes of the systematic review were then used to inform the case study in Chapter Two. The case study investigated the effectiveness of a function-based behavioural sleep intervention to improve sleep for Matthew, a 5-year-old child with CF. Matthew's parents were primarily concerned by his frequent NW and unwanted co-sleeping, as well as EMW. Functional Behaviour Analysis (FBA) revealed Matthew's NW were predominantly maintained by parental presence, and that a lack of physiological sleep pressure also contributed to his sleep problems. The function-based sleep intervention involved three subphases including: (1) sleep/wake rescheduling; (2) a social story and reinforcement; and (3) limiting parental responses to NW. The case study aimed to investigate whether: (1) a behavioural sleep intervention is effective in the treatment of sleep problems in a child with CF; (2) whether any treatment effects are maintained at Short Term Follow Up (STFU); (3) if the selected sleep intervention is acceptable to the parents; (4) whether improving the child's sleep affects the child's daytime behaviour and HRQOL; and (5) whether improving the child's sleep affects the wellbeing of their parents.

Over the course of Matthew's intervention, the frequency and duration of NW and EMW improved, as well as Matthew's ability to settle back to sleep independently following NW. There were no changes in his SOL nor frequency of curtain calls. Although Matthew continued to wake throughout intervention, improvements were considered socially significant to Matthew's father. Furthermore, the intervention resulted in small secondary improvements in Matthew's daytime emotions and behaviours and HRQOL, as well as Matthew father's sleep, although there were no changes in the father's wellbeing and little change in parental relationship satisfaction. The intervention was also reported to be acceptable to Matthew's father.

Caution notwithstanding, the findings of the systematic review and case study collectively suggest that psychosocial sleep interventions may effectively reduce sleep problems in children with CF in the home-setting, and that such improvements may be maintained over time. Furthermore, the findings collectively are suggestive of the safety and suitability of psychosocial sleep interventions for children with CHC. Overall, these findings underscore previous research that suggests that modifiable, behaviourally-based factors contribute to the sleep problems of children with CF (Byars et al, 2020; Canter et al., 2021; Fauroux et al., 2021). Thus the use of FBA within the case study shows that sleep problems of children with CF are not just related to medical/illness factors which cannot be targeted through psychosocial intervention.

The sleep intervention for Matthew included sleep/wake rescheduling, a social story, reinforcement and changes to the parental response which followed NW to address the putative functions for his sleep problems. This lends support to the findings of the review, which showed that psychosocial sleep intervention strategies, including reinforcement and extinction procedures were effective in improving sleep outcomes in children with CHC. Both these strategies were used as part of multicomponent interventions within the reviewed

studies, making it difficult to determine their contribution toward intervention effectiveness, however previous research in typically developing children and children with neurodevelopmental disabilities have found these strategies effective in reducing sleep difficulties (France & Blampied, 2005; Weiskop et al., 2005; Carnett et al., 2020). Specifically, the case study provided support for the use of extinction-based procedures to remove reinforcement (e.g., parent's attention) maintaining sleep problems. For example, the strategy implemented in Subphase 3, which reduced the amount of parental attention Matthew received following NW, resulted in a significant improvement in Matthew's ability to sleep independently. Thus, demonstrating that an extinction-based strategy was effective in reducing problematic NW and co-sleeping in a child with CF. This in line with previous research in typically developing children and children with neurodevelopmental disabilities, as various extinction techniques have been used to reduce children's dependence on parents during sleep onset and throughout the night (Owens et al., 1999; Mindell et al., 2006).

Although no studies within the review specified using sleep/wake rescheduling or social stories, the FBA identified that Matthew's NW were likely occurring due to his inability to self-settle because of his dependence on parental presence and a possible lack of physiological sleep pressure, therefore these strategies were considered important for addressing the maintaining factors of his sleep difficulties. Previous research in children with neurodevelopmental disabilities has found that sleep/wake rescheduling can successfully reduce or eliminate sleep problems, including NW, as this strategy targets the antecedents of sleep problems through increasing sleep pressure (Ford et al., 2021). Thus it was important this strategy was utilised for Matthew as an attempt to address the possible contributing factors to his sleep problems.

It is also possible that the social story (coupled with reinforcement) helped Matthew learn to change his sleep behaviour, suggesting that modeling and providing reinforcement

for appropriate behaviours may be important strategies to use in combination. The use of social stories have successfully been used within typically developing children and children with neurodevelopmental disabilities (Burke et al., 2004; Moore, 2004; Hunter et al., 2021), thus the case study extends their use to show they may also be an effective component of future sleep interventions in children with CF. Overall, these strategies illustrate that targeting the antecedents and consequences of sleep problems in children with CF is important for effectively reducing sleep problems. Thus, the results of the case study add to what is known regarding non-medical interventions for sleep problems in children with CF.

Findings of the systematic review and case study also provide some evidence that secondary benefits may occur following psychosocial sleep interventions. These findings are important as they illustrate the need for researchers and clinicians to routinely assess the sleep of children with CHC, including CF, as addressing sleep problems may also result in improvements in other areas of their lives. Previous research has found associations between sleep problems and daytime impairment for children with CF (Dancey et al., 2002; Bouka et al., 2012; Forte et al., 2015; Íscar-Urrutia et al., 2018; Shakkottai et al., 2018; Tomaszeck et al., 2018; Vandeleur et al., 2018), as well as associations between their sleep problems and impairments in their parents sleep and wellbeing (Sivertsen et al., 2009; McQuillan et al., 2019; Byars et al., 2020). Furthermore, following behavioural sleep interventions, improvements in secondary outcomes have been reported to occur for children with neurodevelopmental disabilities, including in regard to internalising and externalising symptoms (Hunter et al., 2020; McLay et al., 2022). Additionally, for parents of typically developing young children, studies have reported improvements in the parent's sleep and mood following sleep interventions for their child (Mindell et al., 2009b). Thus findings of the systematic review and case study are in line with previous research and provide further support for associations between improved sleep and secondary outcomes. However owing to

the limited nature of the results, further replication within children with CF is required to confidently assert that psychosocial sleep interventions will result in secondary improvements for this population.

Finally, in terms of treatment acceptability and feasibility, the systematic review and case study suggest that psychosocial sleep interventions in general may be feasible and acceptable for families of children with CHC, including CF. It is however important to note that there were only eight studies within the systematic review that assessed social validity, which limits the generalisability of these findings. Further, the interventions in the reviewed studies varied in structure and delivery, including in the duration of intervention (e.g., on-going support versus a one-off session), the frequency and intensity of clinician support, and whether intervention was delivered face-to-face or online. Such factors may differentially impact how acceptable parents find treatment, and it is important that future research considers these factors when designing interventions that meet the family with CF's needs. There are also various child and family factors that may impact treatment acceptability and adherence. For example, the severity of the child's illness and the burden associated with managing their symptoms, as well as the family's available economic and time resources may impact their willingness to engage with interventions, and consequently their perspective of the intervention. Therefore it may be important that future studies consider utilising the same approach within the case study, and investigate the use of staggering function-based treatment components according to the principles of minimal sufficiency. This may ensure adherence and acceptability is optimised.

Strengths and Contributions of the Current Research

The research in this thesis is strengthened by the inclusion of the systematic review. The review enabled the author to identify psychosocial sleep interventions for children with CHC, and appraise the evidence-base for these interventions. The results then informed the

case study in terms of which available methods were likely to be suitable and effective for Matthew. In return, the results of the case study contribute to what is known in the literature regarding the appropriateness and effectiveness of psychosocial sleep interventions for children with CF.

Strengths of the case study included that it is one of the first studies to have investigated the primary and secondary effects of a behavioural sleep intervention in a child with CF. This includes that there has been no research into the use of psychosocial sleep interventions among children with CHC in New Zealand, thus this research enhances national sleep intervention literature. The case study is also one of few studies to have investigated the secondary benefits of sleep interventions on daytime behaviour and HRQOL in children with CF, or other CHC. Furthermore, this is the only study to investigate the secondary benefits of a behavioural sleep intervention with regard to the wellbeing and the relationship satisfaction of parents of a child with CF.

The use of FBA to inform the intervention is also a strength of the case study. FBA enables interventions to be individualised according to the specific needs of the child and their family (Jin et.al., 2013; McLay et.al., 2019). Furthermore, FBA allowed the research team to ensure that Matthew's sleep difficulties had a behavioural-basis and were not the result of medical difficulties (e.g., nocturnal breathing difficulties) which would have required alternative intervention. Previous research has noted the importance of ruling out possible biological or medical causes of sleep problems in children (McLay et al., 2022), which is especially important for children with CF who may suffer from illness-related sleep difficulties which impair their sleep (e.g., respiratory symptoms overnight). Furthermore, as previous research has suggested that 'one size fits all' approaches are not as effective in reducing sleep difficulties as those tailored to the individual (Singh & Zimmerman, 2015;

Spruyt & Curfs, 2015), the use of FBA was important as it ensured the intervention targeted the specific factors underlying sleep difficulties for Matthew.

Another strength of the case study was the single-case research design, which included close monitoring of Matthew's sleep throughout intervention, as it enabled the research team to modify his intervention plan (e.g., the introduction of subphases) in accordance with his progress. Furthermore, the inclusion of STFU was a strength of the case study as it enabled investigation into whether treatment effects were maintained over time. STFU is an important component of research, as it provides a means to determine whether improvements (i.e., changes in behaviours) continue beyond the intervention period, where there is no longer researcher support or involvement.

Directions for Future Research

The findings of Chapters Two and Three highlight a number of potential directions for future research. First, future research examining the effectiveness of psychosocial sleep interventions should aim to include a greater number and diversity of participants with CF, and with CHC in general. This includes participants of different ages, genders, ethnicities, CHC, and different levels of illness severity. Illness severity is an important aspect of CF which can vary immensely (Castellani & Assael, 2017); future research should therefore explore how changes in illness severity can impact intervention, and what specific adaptions may be needed to accommodate differences in severity. Intervention may also need to be tailored to accommodate these different factors. For example, during adolescence there may be many additional factors which impact intervention processes, such as puberty, increasing social pressures and worsening of CF symptoms (Pfeffer et al., 2003; Segal, 2008; Barker & Quittner, 2016b). Therefore FBA may be important for identifying the need for differing techniques between adolescents and young children, such as using CBT-I interventions to address anxiety underpinning insomnia for adolescents (van Deurs et al., 2021). It is therefore

important that future research investigate what components of psychosocial sleep interventions are effective among diverse children with a variety of CHC.

Furthermore, in order to ensure psychosocial sleep interventions are culturally appropriate, research is needed to investigate the types of adaptations that may be required to ensure that assessment and intervention processes are culturally responsive. Within the systematic review, many of the studies were undertaken in western countries, limiting possible understandings of how effective psychosocial sleep interventions are for children with CHC of other ethnicities. Increased marginalisation is associated with decreased health status (Robards et al., 2020); thus, it is important that research assessing the effectiveness of psychosocial sleep interventions not only includes children with varying levels of health, but also children with various cultural identities. For example, within Māori culture, health can be understood through various models including Te Whare Tapa Whā (Durie, 1994). Te Whare Tapa Whā emphasises the importance of considering Taha tinana (physical health), Taha wairua (spiritual health), Taha whānau (family health) and Taha hinengaro (mental health) within healthcare, as when one of these dimensions is hindered, a person's overall health may become 'unbalanced'. Therefore within psychosocial sleep interventions for Māori children, it may be especially important to consider other areas of functioning (e.g., spirituality) to ensure that interventions are responsive to the child and family's sleep and cultural needs. Thus, further research is required to gather information on the cultural appropriateness and acceptability of such interventions.

As discussed in Chapter Three, it is also important that future research includes objective measures (e.g., actigraphy, videosomnography) of sleep, in addition to parent-reported measures (e.g., sleep diaries). A combination of measures will enable stronger conclusions to be drawn regarding the effectiveness of interventions (Moore et al., 2017). For example, although sleep diaries provide valuable information regarding parents' perspectives

of their child's sleep, and allow for reporting of information that cannot be captured through objective measures (e.g., things that have happened in the day which may contribute to sleep problems), the use of objective measures provides another source of data to enable parent-reported information to be triangulated.

The use of objective and subjective measures may also assist in intervention planning and monitoring to ensure clinicians have an accurate and comprehensive understanding of a child's sleep habits and behaviour. For example, within the case study, it is unclear whether there were nights where Matthew resettled independently following NW without signalling to his parents that he was awake (i.e., a quiet awakening), or if he did not wake at all. Videosomnography would have provided valuable insight into what may have happened during the night that his parents may have been unaware of, and therefore were unable to report.

Finally, because illness exacerbations are a significant factor which may impact intervention effectiveness for children with CF (Shakkottai et al., 2018), the inclusion of illness-related measures may be beneficial to future studies. For example, regularly measuring Forced Expiratory Volume (FEV1), via in-home spirometry tests, may inform clinicians about the health of children with CF during assessment and intervention. This would enable clinicians to monitor improvements in FEV1 in relation to sleep, and to monitor illness symptoms to ensure that the physical health of the child is being prioritised. The use of such measures before, during, and after intervention may allow inferences to be made regarding the effects of improved sleep on the lung functioning of the child. Previous studies have reported that despite children with CF being considered clinically stable, objective measures of sleep have found sleep quality, SE, TST, frequency of NW, and WASO, to be related to illness severity as measured by FEV1 (Vandeleur et al., 2017). Thus, this is potentially an important avenue for future research to investigate.

Clinical Implications

The current research provides insight into the effectiveness and acceptability of psychosocial sleep interventions for children with CHC, including CF. The review and the case study highlight the importance of assessing behavioural and psychological factors underpinning sleep difficulties in children with CHC, as these difficulties can be amenable to psychosocial sleep interventions. Previous research has illustrated that although children with CF experience high rates of sleep difficulties, sleep difficulties are not often discussed within routine care CF visits (Canter et al., 2021). It is important that screening for sleep difficulties are incorporated within standard CF care, and that families are educated about the importance of sleep for their child with CF. This may include psychoeducation and assessment of difficulties relating to the initiation and maintenance of sleep, including the presence of sleep disordered breathing, which is common for children with CF (Shakkottai et al., 2022).

An important clinical implication of the case study was that FBA enabled the intervention to be tailored according to the presenting sleep difficulties and putative functions for Matthew as an individual. The intervention was then designed to address these specific underlying factors. Previous research has indicated that adolescents with CF are open to behavioural sleep interventions that are flexible and specific to their needs (Canter et al., 2021). Thus, the use of FBA may be important to informing intervention within practice to effectively target the underlying causes of sleep problems, while ensuring family needs are met in an efficient way that does not exceed the clinician's and family's resources.

Importantly, the case study methodology involved regular contact between the clinician and family throughout the intervention process. This ensured that sufficient levels of clinical input were maintained throughout intervention, which allowed any possible barriers to treatment adherence to be resolved as they arose. It also allowed rapport to be built between the clinician and family which may have contributed to high treatment acceptability.

The review by Lunsford-Avery et al. (2021) which was discussed in Chapter Two, found that interventions were more likely to be effective if delivered by an individual, rather than solely through printed materials. Thus, the in-person mode of delivery within the case study may have also contributed to the effectiveness of the intervention in improving Matthew's sleep.

Conclusion

The overarching aim of this thesis was to investigate the effectiveness of psychosocial sleep interventions for children with CF. Collectively, findings from the review and case study suggest that psychosocial sleep interventions may be effective and appropriate for use in children with CHC including CF. However, given that there was only one child participant in the case study, and only five reviewed studies that included children with respiratory conditions (and none with CF), the strength of conclusions regarding the effectiveness of psychosocial sleep interventions in children with CF is limited. Nevertheless, the results of this research provides impetus for future studies to further investigate how to decrease the burden of sleep problems for children with CF and their families. Future research may attempt to address some of the previously identified limitations by including more participants with diverse presentations and utilising objective as well as subjective measures. Further research is also needed to better understand the nature of the bidirectional relationship between sleep and illness-related symptoms in children with CF.

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Appendix A: Parent Consent Form

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

CONSENT FORM FOR PARENTS/ CAREGIVERS

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

☐ I wish to participate in the project, *"An investigation into the effectiveness of treatments for sleep disturbance in children with Cystic Fibrosis".*

☐ I have read and been given a full explanation of this project and have had the opportunity to ask questions.

☐ I understand what will be required of myself and my child/the child in my care during this project

☐ I understand that the investigators do not foresee any potential risks to me or my child as a result of participating in this study. However, if the intervention results in an increase in family stress, the staff working with us will provide support.

☐ I understand that all information about my family will be treated as confidential unless there is concern about anyone's safety. In this case my clinician will need to speak to someone else to ensure the safety risk is removed. No findings that could identify me or my child will be published

☐ I understand that the findings of this study may be published in a research journal or at a conference and that the anonymity of my child and I will be maintained

☐ I understand that participation in this project is voluntary and that I can withdraw my child or he/she can withdraw from the project at any time without repercussions. I can also withdraw any data that has been collected at any time prior to the publication of that data

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

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

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

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

Name: _____

Date: _____

Signature: _____

Others I consent to implementing intervention:

Name:_____

Name:_____

Name:_____

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

Please return this form to Laurie McLay.

Appendix B: Information Sheet for Parents

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

Information for Parents/Caregivers

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

Dear Parent/ Caregiver,

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

We would like you and your child with Cystic Fibrosis 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 Cystic Fibrosis. Treatment can include a range of strategies, including both non-traditional approaches (such as white noise) and behavioural interventions. These approaches have been designed to minimise stress as much as possible for the parents and children using them. We are also interested in parents' and children's experiences in using the treatments and any changes to their lives, or their child's lives, which result.

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

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

When we have established an understanding of your child's sleep behaviour, we will work with you to develop sleep-related goals for your child. This will involve a second treatment-planning session which will last 1-1 ½ hours.

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

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

For the purpose of this project, myself, Laurie McLay (lead investigator) and a registered psychologist will be working closely with you to conduct the necessary assessments and formulate interventions. Emma Woodford, a PhD student who also works as a part of our sleep team, along with a Research Assistant 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 C: Standard Parent-Report Sleep Diary

		Monday	Tuesday	Wednesday	Thursday	Friday	Saturday	Sunday
Night-time sleep	Setting (where he fall asleep)							
	Who put him to bed							
	Time he was put to bed							
	Frequency of curtain calls Describe each curtain call Parent's responses to each curtain call							
	Best estimate of time asleep							
	Time bedroom door closed							
Night waking	Time and duration of waking							
	Describe behaviour while awake							
	Parent's responses							
Morning	Time awake in the morning							