SLEEP PROBLEMS IN CHILDREN WITH NEURODEVELOPMENTAL DISORDERS: BARRIERS AND FACILITATORS TO TREATMENT AND DEVELOPMENT OF AN ONLINE INTERVENTION FOR INSOMNIA

by

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Abstract

Neurodevelopmental disorders (NDDs) emerge early in life and are associated with functional impairment for children and their families. Amongst children with NDDs, sleep problems are highly prevalent and can have widespread negative effects on children's health, functioning, and quality of life. However, there are few established evidence-based sleep interventions and access to these is limited. This three-study dissertation aimed to lay the groundwork for developing a transdiagnostic eHealth parent-implemented intervention for children with four NDDs (Autism Spectrum Disorder [ASD], Attention-Deficit / Hyperactivity Disorder [ADHD], Cerebral Palsy [CP], and Fetal Alcohol Spectrum Disorder [FASD]). Study 1 is a systematic review of online parent-implemented interventions for NDD symptoms and other behaviour problems in children with NDDs. Twelve interventions were identified, for ASD (*n* = 8), ADHD (*n* = 3), and FASD (*n* = 1). Despite promising evidence for the effectiveness of online parent-implemented interventions and their potential to resolve treatment access problems, no interventions currently qualify as evidence-based and more large-scale trials are required. Study 2 qualitatively explored barriers and facilitators to sleep treatment access, implementation and provision experienced by parents and health care professionals (HCPs) of children with ASD, ADHD, CP and FASD via focus groups / interviews. Similar themes emerged across all NDDs. Key barriers reported by parents (*n* = 43) and HCPs (*n* = 44) included lack of knowledge / awareness of sleep problems and their treatment in NDDs, limited access to treatment, the demanding nature of treatments, and parent factors (e.g., exhaustion). Key facilitators included education, support, behavioural treatment approaches, and the ability to modify treatments to account for NDD symptoms. In Study 3, parents of children with ASD, ADHD, CP, or FASD (*n* = 20) implemented an eHealth parent-implemented insomnia intervention designed for typically developing children, and evaluated the intervention's usability, accessibility, and feasibility. Parents found it usable, acceptable, and feasible, and suggested modifications to make the intervention more useful for children with NDDs. Overall, these studies demonstrate the need for an accessible sleep intervention, and support a transdiagnostic approach to treating sleep problems in children with NDDs.

List of Abbreviations Used

ADHD = Attention-Deficit / Hyperactivity Disorder

ASD = Autism Spectrum Disorder

BCBA = Board Certified Behaviour Analyst

BIQ = Behavioural Insomnia Questionnaire (measure)

BNBD = *Better Nights, Better Days* (name of intervention)

BNBD-NDD = *Better Nights, Better Days for Children with Neurodevelopmental Disorders* (name of intervention)

CP = Cerebral Palsy

DNC = did not complete

ES-UQ = End of Session Usability Questionnaire (measure)

FASD = Fetal Alcohol Spectrum Disorder

HCP = health care professional

N; *n* = sample size

NDD(s) = neurodevelopmental disorder(s)

O-UQ = Overall Usability Questionnaire (measure)

PICOS = Population, intervention, comparison, outcome, study design (model)

PP = pre/post study

PRISMA = Preferred Reporting Items for Systematic Reviews and Meta-Analyses

PSQ = Pediatric Sleep Questionnaire (measure)

RCT = randomized controlled trial

SC = single-case design

SD = standard deviation

SILS = Single Item Literacy Scale (measure)

TD = typically developing

WLC = wait list control

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Chapter 1: Introduction

Brief Overview

Sleep problems are highly prevalent in children with neurodevelopmental disorders (NDDs) and can potentially have a negative impact on children and families. As such, there is a great need to develop accessible, effective interventions for sleep problems in children with NDDs. Research on sleep treatment accessibility, uptake, and provision from the perspectives of both families and health care professionals (HCPs) is limited.

In this dissertation, I first review prior research on online parent-implemented interventions, a mode of treatment delivery that could increase treatment accessibility. Next, I explore barriers and facilitators to sleep treatment access and use as perceived by parents of children with NDDs and their HCPs. Finally, I evaluate whether parents of children with NDDs perceive an eHealth parent-implemented behavioural intervention originally designed for insomnia in typically developing (TD) children to be usable and acceptable, and assess what changes are needed to modify the intervention to suit the needs of children with NDDs and their parents. This dissertation focuses on four NDDs that represent a range of symptoms and functional impairments: Autism Spectrum Disorder (ASD), Attention-Deficit / Hyperactivity Disorder (ADHD), Cerebral Palsy (CP), and Fetal Alcohol Spectrum Disorder (FASD) (collectively referred to throughout this dissertation as NDDs). These four NDDs were selected because they are the focus of NeuroDevNet (now Kids Brain Health Network), the funding entity that supported the development of the eHealth insomnia intervention, entitled *Better Nights, Better Days for Children with Neurodevelopmental Disorders* (BNBD-NDD).

Before presenting these studies, it is important to provide some information about the four NDDs of interest, sleep problems in children with NDDs, barriers to accessing sleep treatment, and potential methods of overcoming such barriers. I will conclude this chapter by reviewing the rationale and objectives of this dissertation.

Neurodevelopmental Disorders in Children

Neurodevelopmental disorders are a class of disorders that emerge early in life and are linked to disturbances in central nervous system functioning, which can manifest as impairments in cognition, communication, motor skills, and/or behaviour that vary in severity (American Psychiatric Association, 2013; Reiss, 2009). They are associated with functional impairment in personal, social, occupational, and / or academic areas (American Psychiatric Association, 2013). The etiology of NDDs is complex and involves both genetic and environmental components (American Psychiatric Association, 2013). Neurodevelopmental disorders frequently co-occur or share symptoms across diagnoses; examples of NDDs based on the DSM-5 include ASD, ADHD, intellectual disability, specific learning disorders, and communication and language disorders, as well as genetic syndromal NDDs such as Fragile X syndrome, medical conditions such as CP, and conditions associated with environmental factors such as FASD (American Psychiatric Association, 2013). A brief description of the four NDDs studied in this dissertation follows.

Autism Spectrum Disorder (ASD) is characterized by persistent deficits in social communication and interaction, and restricted, repetitive behaviours, interests, and activities (American Psychiatric Association, 2013). In Canada the prevalence of ASD is currently 1 in 66 children (Public Health Agency of Canada, 2018). Attention-Deficit / Hyperactivity Disorder (ADHD) is characterized by a persistent pattern of inattention and / or hyperactivity-impulsivity, present before twelve years of age and occurring in more than one setting, that impairs children's functioning or development. With a prevalence rate of between 5 and 7% (Wilcutt, 2012), ADHD is reported to be the most common childhood NDD. Cerebral Palsy (CP) constitutes a group of motor disorders that affect movement and posture and are often accompanied by concurrent seizure disorders and/or impairments in cognition, communication, perception, behaviour, and sensation (Rosenbaum, Paneth, Leviton, Goldstein, & Bax, 2007). With an overall prevalence of 2.11 per 1000 live births, CP is the most common cause of childhood physical impairment (Oskoui, Coutinho, Dykeman, Jette, & Pringsheim, 2013). Fetal Alcohol Spectrum Disorder (FASD) refers to a spectrum of disorders linked to adverse health effects stemming from prenatal exposure to alcohol, with prevalence estimated at 8 in 1000 persons (Chudley et al., 2005; Lange et al., 2017). Because alcohol can affect any organ or system during development, children with FASD may experience a wide range of deficits in cognitive, behavioural, emotional, and adaptive functioning, as well as a range of comorbid conditions (Chudley et al., 2005).

Sleep Problems in Children with NDDs

Sleep problems occur in 50 – 90% of children with NDDs (Chen, Olson, Picciano, Starr, & Owens, 2012; Cortese, Faraone, Konofal, & Lecendreux, 2009; Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008; Romeo et al., 2014). Concerningly, sleep problems can negatively affect many aspects of functioning in children with NDDs and interfere with the effectiveness of interventions for NDD symptoms (Goldman, McGrew, Johnson, Richdale, Clemons, & Malow, 2011; Tudor, Hoffman, & Sweeney, 2012; Vriend, Corkum, Moon & Smith, 2011). In children with NDDs, the most commonly experienced sleep problems are collectively referred to as insomnia, including difficulty falling asleep, difficulty staying asleep, and nighttime or early morning awakenings (American Academy of Sleep Medicine, 2014; American Psychiatric Association, 2013). Given the high rate of sleep problems in children with NDDs and the potential impacts on functioning, the development of effective interventions for sleep problems is critical to maximize quality of life in these children and their families.

Sleep problems in children with NDDs may stem from combinations of multiple factors, including: neurobiological factors, primary sleep disorders, comorbid neurological / medical / psychiatric disorders, medication use, and behavioural factors that include child and family variables (Corkum, Weiss, Tan-MacNeill, & Davidson, 2014; Reynolds & Malow, 2011; Stores, 2016). Neurobiological factors may include disruption of the neurotransmitters that promote sleep and establish a regular sleep-wake cycle (i.e., gamma aminobutyric acid, serotonin, and melatonin). Primary sleep disorders include sleep-disordered breathing (e.g., obstructive sleep apnea), parasomnias (e.g., sleepwalking, night terrors, confusional arousals, frequent nightmares), sleep-related movement disorders (e.g., restless legs syndrome), and circadian rhythm disorders. Comorbid neurological, medical, and psychiatric disorders, including epilepsy, gastrointestinal disease (e.g., reflux), eczema, allergies, recurrent infections, mental health problems such as anxiety or depression, and primary enuresis or encopresis can also impact sleep. Use of medications for comorbid neurological, medical, and mental health disorders (e.g., stimulant medications for ADHD) can also disrupt sleep.

Insomnia is often described as a primarily behaviourally based sleep disorder, and behavioural factors are thought to play a significant role in causing insomnia in children with NDDs (Corkum et al., 2014; Reynolds & Malow, 2011). These behavioural factors can include individual child factors as well as family variables. For example, because sleep occurs in a complex psychosocial context, family variables such as household routines, parental mental health, family composition, and work and school schedules may also affect children's sleep. Parenting styles, parental expectations, and family sleep hygiene practices can also play a role. Individual child and behavioural factors include temperament, unhealthy sleep routines or habits, inflexibility or rigidity (particularly with regard to habits and routines), hypersensitivity to environmental stimuli (e.g., temperature in bedroom, level of light), hyperarousal, difficulty with self-regulation, repetitive thoughts or behaviour that interfere with settling before bedtime, and lack of ability to benefit from social or communicative cues about sleep (Corkum et al., 2014; Jan et al., 2008; Reynolds & Malow, 2011).

Impact of Sleep Problems on Children with NDDs

Across NDDs, sleep problems are correlated with increased NDD symptoms. For example, in children with ASD, sleep problems are associated with increased severity of symptoms such as stereotyped behaviours, communication problems, social interaction problems, and sensory differences (Goldman et al., 2011; Hollway, Aman, & Butter, 2013; Mayes, Calhoun, Bixler, & Vgontzas, 2009; Segawa, Katoh, Katoh, & Nomura, 1992; Taylor, Schreck, & Mulick, 2012; Tudor, Hoffman, & Sweeney, 2012). In children with ADHD, sleep problems are correlated with increased hyperactivity and inattention and poorer cognitive functioning, especially in executive functioning skills (Corkum et al., 1999; Konofal, Lecendreux, & Cortese, 2010; Mayes et al., 2009; Moreau, Rouleau, & Morin, 2013). In children with FASD, sleep problems are linked to externalizing and internalizing behaviour problems as well as emotion regulation difficulties (Chen et al., 2012; Ipsiroglu, McKellin, Carey, & Loock, 2013; Jan et al., 2010). In children with CP, sleep problems are associated with physiological NDD symptoms, including greater motor impairment, increased frequency of pain, and increased involuntary movement (Newman, O'Regan, & Hensey, 2006; Romeo et al., 2014; Simard-Tremblay, Constantin, Gruber, Brouillette, & Shevell, 2011).

Sleep problems can also negatively affect the overall cognitive, emotional, and daytime functioning of children with NDDs (Chen et al., 2012; Corkum et al., 1999; Hollway, Aman, & Butter, 2013; Ipsiroglu, McKellin, Carey, & Loock, 2013; Jan et al., 2010; Newman et al., 2006; Owens, 2009; Sandella, O'Brien, Shank, & Warchausky, 2011; Schreck, Mulick, & Smith, 2004; Taylor, Schreck, & Mulick, 2012; Wengel, Hanlon-Dearman, & Fjeldsted, 2011). For example, sleep problems in children with ASD are associated with poorer adaptive skills such as those related to hygiene, toileting, and eating (Taylor et al., 2012). For both CP and FASD, sleep problems are associated with cognitive impairment (Romeo et al., 2014; Jan et al., 2010). Clearly, insomnia significantly affects the functioning of children and has wide-ranging effects on NDD symptoms and associated challenges.

Impact of Children’s Sleep Problems on Families

Sleep problems can also significantly affect the entire family, given the psychosocial context of children’s sleep problems (Bernier et al., 2013; El-Sheik & Kelly, 2017; Simard-Tremblay et al., 2011). Most research on familial impact of children’s sleep problems has focused on parents, rather than siblings. Parents of children with NDDs and sleep problems may experience poor sleep themselves (Hoffman, 2008), be vulnerable to lower parenting self-efficacy (Hastings & Brown, 2002), and experience higher levels of stress (Doo & Wing, 2005; Hoffman et al., 2009). It is not clear whether the relationship between parent stress and child sleep problems is bidirectional, but certainly parent stress has been shown to affect parenting behaviour, which could further affect parents’ ability to manage their children’s sleep problems (Hastings, 2002). Parents of children with ASD perceive their children’s insomnia as increasing parental anxiety and stress, adversely affecting the relationship between partners / spouses, and having negative financial consequences (Kirkpatrick, Gilroy, & Leader, 2019). Parents of children with ADHD who have moderate to severe sleep problems are three times more likely to have elevated stress levels, as well as higher risk of depression and anxiety (Sung, Hiscock, Sciberras, & Efron 2008). Sleep problems in children with FASD are associated with caregiver burden and poorer quality of life, two outcomes that are particularly salient given the already high levels of stress and psychological distress amongst FASD caregivers (Hanlon-Dearman, Chen, & Olson, 2018; Ipsiroglu et al., 2013). Similarly, research on parents of children with disabilities and sleep problems, including CP, shows that parents feel more stressed and irritable, get less sleep, and perceive a negative impact on their own daytime lives and work (Simard-Tremblay et al., 2011).

Treatment of Sleep Problems in Children with NDDs: What Do We Know from the Literature on Typically Developing Children?

Most research on evidence-based intervention for pediatric behavioural sleep problems has been conducted with TD children (Melzter & Mindell, 2014; Zhou & Owens, 2016). Behavioural interventions are the first-line, recommended treatment for sleep problems, and are based on the underlying principle that healthy sleep is a learned behaviour (Corkum et al., 2014; Mindell, Kuhn, Lewin, Meltzer, & Sadeh, 2006). It is recommended that children be screened to determine whether sleep problems are caused by any sleep disorders or medical factors (for example, obstructive sleep apnea), which should be treated first by an appropriate clinician (Corkum et al., 2014; Malow et al., 2012). The following stepped or staged approach is recommended for behavioural sleep problems, such that each step progresses to a more intensive intervention (based on Corkum et al., 2014; Malow et al., 2012; Mindell et al., 2006 – see these articles for a full review of behavioural interventions for sleep problems, which is beyond the scope of this chapter).

Step 1: Parental Beliefs and Sleep Education

Parents' knowledge and beliefs about sleep can influence their use of sleep strategies (Bessey, Coulombe, Smith, & Corkum, 2013). As such, psychoeducation about the causes, symptoms, consequences, and treatment of sleep problems is recommended. It is especially important to ensure parents know the importance of consistency in treating sleep problems and establishing a regular sleep schedule.

Step 2: Sleep Hygiene and Bedtime Routines

Developing good bedtime routines and practicing healthy sleep habits is a critical piece of sleep treatment. Key practices include: providing or creating an optimal sleep environment (e.g., ensuring temperature, light, and sound are conducive to sleep), sleep scheduling (ensuring consistent bedtimes and waketimes), sleep practice (i.e., ensuring that children are taught how to fall asleep independently from their parents), and physiological sleep-promoting factors (e.g., ensuring children receive enough exercise during the day, avoiding caffeinated foods before bedtime, eliminating use of electronic devices before bedtime or in the bedroom). Other practices include incorporating positivity and relaxation into the bedtime routine, meeting children's emotional needs during the day, and ensuring that bedtimes and sleep duration are age-appropriate.

Step 3: Specific Behavioural Strategies

Effective strategies for improving sleep in TD children include extinction / graduated extinction, faded bedtime with sleep restriction, cognitive strategies, and reward or reinforcement programs (Zhou & Owens, 2016). Extinction is effective for treating difficulties with falling asleep independently (e.g., child cries out at night, requires parents to be in bedroom to fall asleep, or co-sleeps). Children are taught to self-soothe and fall asleep independently as parental presence is reduced and eliminated from the bedroom. Faded bedtime with sleep restriction techniques is useful for addressing difficulties with falling and staying asleep. Parents identify a target bedtime, and delay or fade bedtime over a period of days or weeks until the target bedtime is achieved and the child falls asleep quickly once in bed. To help the child become sleepier and fall asleep faster, sleep quantity can be restricted. Cognitive strategies such as coping / relaxation skills and cognitive restructuring of unhelpful beliefs about sleep can be used for children and parents, especially children who experience anxiety about sleep and parents who are coping with negative thoughts about sleep. Finally, reward and reinforcement programs, such as a token economy or a bedtime pass program, can be used to increase desired behaviour (e.g., staying in bed) and decrease unwanted behaviour (e.g., calling out to parents); these methods can be especially helpful for bedtime disturbances.

Step 4: Pharmacological Intervention with Behavioural Strategies

Pharmacological intervention should not be the first step in treatment, but should only be considered if behavioural strategies do not resolve sleep problems, and used in conjunction with behavioural strategies (Barrett, Tracy, & Giaroli, 2013; Blackmer & Feinstein, 2016; Corkum et al., 2014; Johnson & Malow, 2008; Malow et al., 2012). Currently, all medication use for sleep treatment in children is off-label (Bruni et al., 2018), and a consensus statement on pharmacological management of sleep in children and adolescents concluded that more research on safety and efficacy was required (Mindell et al., 2006). In spite of this, pharmacological management of children's sleep problems is widespread. For example, a survey of community-based American paediatricians found that 75% recommended over-the-counter sleep aids to treat sleep in children, 50% prescribed medication to manage pediatric insomnia, and moreover, that paediatricians reported using medications to treat sleep more frequently for children with NDDs than TD children (Owens, Rosen, & Mindell, 2003). Another study found that at least one medication was prescribed for 46% of all children with ASD who were diagnosed with a sleep disorder, with melatonin being most common (Malow et al., 2016). Melatonin use is extremely common in NDD populations (Bruni et al., 2015), possibly because it is available over-the-counter in many jurisdictions (Cummings, 2012). Melatonin has been found to be effective in improving sleep onset latency and total sleep quantity with minimal side effects in children with ASD and/or ADHD (Abdelgadir, Gordon, & Akobeng, 2018; Gringras et al., 2012; Hoebert, van der Jeijden, van Geijlswijk, & Smits, 2009; Hollway & Aman, 2011; Maras et al., 2018; McDonagh, Holmes, & Hsu, 2019). However, more research on safety, efficacy, and dosage is needed (Bruni et al., 2015; Cummings, 2012). A full review of medication for sleep problems is beyond the scope of this chapter, but the reader is referred to reviews by McDonagh and colleagues (2019), Blackmer and Feinstein (2016), and Hollway and Aman (2011).

Translating Sleep Intervention Recommendations from TD to NDD

Overall, the literature recommends that the same behavioural strategies and approaches for sleep problems in TD children be used for children with NDDs (Blackmer & Feinstein, 2016; Bruni et al., 2018; Grigg-Damberger & Ralls, 2013; Robinson-Shelton & Malow, 2016). These recommendations are consistently made for children with ASD (Cortese et al., 2020; Johnson & Malow, 2008; Malow et al., 2012), ADHD (Hvolby, 2015), CP (Brown et al., 2013; Simard-Tremblay et al., 2011), and FASD (Hanlon-Dearman et al., 2018; Jan, 2010; Wengel et al., 2011). The most common behavioural interventions used to treat sleep problems in children with ASD, ADHD, CP, and FASD are implementation of healthy sleep practices, reinforcement, graduated extinction, and faded bedtime (Rigney et al., 2018). In a systematic review, Rigney and colleagues (2018) found that these strategies were similar to those used with TD children and were used across NDD diagnoses, suggesting that a transdiagnostic behavioural sleep intervention approach may be warranted. This is consistent with research that suggests sleep problems are transdiagnostic (occurring across multiple disorders) and that transdiagnostic intervention may be an effective way to treat insomnia in adults and children (Harvey, Murray, Chandler, & Soehner, 2011; Harvey, 2016).

Although evidence suggests that core behavioural strategies can be used transdiagnostically, modifications can be made to accommodate the needs and symptoms of children with NDDs if necessary (Ali et al., 2018; Jan et al., 2008; Rigney et al., 2018). These may include adjusting the pace and duration of treatment, using specific forms of cues (e.g., visual schedules), tailoring behavioural reinforcers to children's interests, and accommodating children's physical disabilities, sensory sensitivities, and attention problems (Jan et al., 2008). Disorder-specific modifications may include addressing the impact of ADHD symptom medication (i.e., stimulants) and adjusting the dose as necessary (Corkum, Panton, Ironside, MacPherson, & Williams, 2008; Hvolby, 2015); paying particular attention to sensory processing difficulties and sensory sensitivities, combined with impaired understanding of environmental cues, in children with FASD and ASD (Jan et al., 2010); screening for attachment and trauma concerns in children with FASD (Hanlon-Dearman et al., 2018; Wengel et al., 2010); and for children with CP, incorporating physical interventions such as massage (Brown et al., 2013) and using pharmacological treatments when behavioural and environmental interventions are not effective (Simard-Tremblay et al., 2011).

Barriers to Accessing and Using Sleep Interventions

Although behavioural interventions are recommended as the first-line evidence-based treatment for sleep problems in children with NDDs, use of pharmacological interventions (e.g., melatonin) and complementary and alternative medicine treatments are far more common and more easily accessible (Cohen et al., 2018; France, McLay, Hunter, & France, 2018; Malow et al., 2016). Evidence suggests limited uptake of behavioural interventions for sleep by families of children with NDDs (Robinson & Richdale, 2004). This suggests that there are barriers to widespread use of behavioural interventions. Two barriers may play a major role: families' lack of access to treatment, and an overall lack of interventions specifically for children with sleep problems and NDDs.

Lack of Access to Treatment

No studies have directly examined barriers to accessing sleep interventions for children with NDDs. However, research has demonstrated that families of children with NDDs in the United States have considerable difficulty accessing evidence-based interventions in general, including access to screening, early intervention, and mental health services, due to lack of coordination of health care services (Johnson & DeLeon, 2016). Others barriers to access can include living in remote and underserved areas, experiencing long wait times to access specialists and treatments, and having difficulties with transportation to appointments (for example, challenges bringing children with behavioural problems and difficulty dealing with transitions into clinic settings; Thomas, Ellis, McLaurin, Daniels, & Morrisey, 2007).

Difficulties with accessing treatment are also linked to health care providers. Research suggests that sleep problems are under-diagnosed in the pediatric population (Meltzer, Johnson, Crosette, Ramos & Mindell, 2010). Front-line Canadian and American HCPs are not generally well-trained in providing sleep interventions (Boerner et al., 2014; Bruni et al., 2004; Faruqui, Khubchandani, Price, Bolyard, & Reddy, 2011; Owens, 2001), nor do they receive training to address sleep problems in special populations such as children with NDDs. For example, a study surveying pediatric medical residency programs across 10 countries showed that the average time spent on sleep education was 4.4 hours, with nearly a quarter of programs offering no sleep education at all (Mindell et al., 2013). Finally, lack of time, lack of funding, and lack of resources and materials can also make it difficult for HCPs to offer or provide sleep treatment, making it harder for parents to access treatment (Boerner et al., 2014; Honaker & Meltzer, 2016).

Lack of Available Programs

The development of effective behavioural sleep interventions for children is a major focus of current research, and several randomized controlled trials (RCTs) have been conducted within the past decade (e.g., Adkins et al., 2012; Corkum et al., 2016; Hiscock, Sciberras, & Mensah, 2015; Papadopoulos et al., 2019). However, few interventions or resources are widely available outside research studies. For example, there is only one published practice guideline for treating sleep problems in children with NDDs: the Autism Treatment Network practice pathway for children with ASD (Malow et al., 2012). For parents, the North American advocacy organization Autism Speaks has developed three "toolkits" focused on sleep strategies, including one for children, one for teens, and a guide to melatonin and sleep (https://www.autismspeaks.org/sleep). Several books for parents have also been published, including two for parents of children with ASD – *Solving Sleep Problems in Children with Autism Spectrum Disorders: A Guide for Frazzled Parents* (Katz & Malow, 2014), and *Sleep Well on the Autism Spectrum* (Aitken, 2014) – and one for parents of children with special needs, *Sleep Better! A Guide to Improving Sleep for Children with Special Needs* (Durand, 2013). Unfortunately, to our knowledge, no such specific resources exist for ADHD, CP, and FASD. Indeed, most behavioural sleep intervention research focuses on children with ASD and / or ADHD (Rigney et al., 2018), with limited research on treatment for children with CP and FASD (Brown et al., 2013; Ipsiroglu et al., 2013).

Given these barriers, it is not surprising that use of pharmacological interventions and complementary and alternative treatment to address sleep problems is widespread in the absence of evidence (Blackmer & Feinstein, 2016; France et al., 2018). Clearly, development of more evidence-based sleep interventions for children with NDDs is needed. However, it is first important to consider how to increase the accessibility of interventions. Two potential methods to do so include using the internet to deliver interventions remotely, and teaching parents to implement interventions themselves.

Increasing Accessibility of Evidence-Based Intervention: Using the Internet and Teaching Parents to Provide Treatment

Online Delivery of Interventions

Remote delivery of interventions can help to increase treatment accessibility. For example, telehealth interventions involve health care professionals or paraprofessionals delivering interventions to families in real-time, via telephone or web-based videoconferencing, and can offer families increased access, convenience, privacy, and ability to receive services remotely (Lingley-Pottie & McGrath, 2013). However, telehealth interventions still require considerable time and involvement of clinicians or paraprofessionals.

In contrast, eHealth interventions are a type of online (i.e., internet- or web-based) intervention defined as “applications of technology that seek to either improve a client’s understanding of health information or use technology as a surrogate for the clinician in a treatment delivery” (Cushing & Steele, 2010, p. 937). Because eHealth interventions are primarily self-directed and require less clinician time than in-person or telehealth interventions, they may be even more cost-effective and accessible (Rapoff, 2013). Over the last decade, online evidence-based interventions have been recommended as a way of addressing gaps in child and adolescent mental health service provision and increasing treatment accessibility and quality (see Comer & Myers, 2016; Rapoff, 2013; Siemer, Fogel, & Van Voorhees, 2011). Reviews of pediatric online interventions have highlighted their potential effectiveness for improving health, behavioural, and mental health outcomes for children and adolescents (Cushing & Steele, 2010; Siemer et al., 2011). I will use the terms eHealth intervention and online intervention interchangeably throughout this thesis.

Teaching Parents to Implement Interventions

Another way to increase treatment access may be to teach parents how to provide treatment to their children. Parent-implemented interventions aim to teach parents strategies to effect behavioural change in their children (i.e., teaching new functional skills, reducing maladaptive behaviours), and have a long history in NDD research (e.g., Schopler & Reichler, 1971). Not only can parent-implemented interventions reduce the need for HCPs to be directly involved in the delivery of treatment (thus increasing treatment access and reducing healthcare system burden), but they can empower parents with greater knowledge, confidence, and skills (Bearss, Burrell, Stewart, & Scahill, 2015). Parent-implemented interventions are effective for improving core NDD symptoms in children with ASD (Althoff, Dammann, Hope, & Ausderau, 2010; Oono et al., 2018) and ADHD (Evans, Owens, Wymbs, & Ray, 2018), as well as for improving communication and feeding behaviours in children with CP (Whittingham, Wee, & Boyd, 2011). For FASD, interventions that teach parents how to change children's maladaptive behaviours are recommended (Paley & O'Connor, 2009).

Combining "Online" with "Parent-Implemented": Online Parent-Implemented Interventions

Unfortunately, parent-implemented interventions are under-utilized in community settings (particularly non-urban / non-metropolitan settings; Thomas et al., 2007), and require considerable resources for training, which may be challenging due to families' schedules and limited HCP resources (Vismara, Young, & Rogers, 2012). Online parent-implemented interventions may be one way to increase accessibility and reduce burden on the healthcare system (Breitenstein, Gross, & Christophersen, 2014; Meadan & Daczewitz, 2015; Nieuwboer, Fukkink, & Hermanns, 2013; Rapoff, 2013). When considered within a "stepped-care model" of healthcare delivery (Espie, 2009), an online parent-implemented intervention may represent an ideal first- or entry-level treatment. Such an intervention can be accessible, convenient, low-cost, reach a large volume of patients, and require less HCP time than an in-person or telehealth intervention (Bower & Gilbody, 2005; Rapoff, 2013). Online interventions may also be interesting and acceptable to parents, who already use the internet to answer their healthcare questions (Di Pietro, Whitely, Mizgalewicz, & Illes, 2013; Thorndike, 2009). Currently, literature on online parent-implemented interventions for children with NDDs is limited, with most studies focusing on parents of children with ASD. Preliminary evidence suggests that online parent-implemented interventions for ASD can improve children's social communication and imitation skills, as well as parent knowledge (Meadan & Daczewitz, 2015). More research is needed on the effectiveness of eHealth parent-implemented interventions for a range of NDD diagnoses. The limited research available about online parent-implemented interventions for children with a range of NDDs suggests a need for a systematic review to gather and evaluate available evidence.

Interestingly, research has suggested that online delivery may be particularly well-suited to pediatric insomnia interventions (Owens, 2014). Considerable evidence has already shown that eHealth interventions are effective for insomnia in TD adults (Espie, Hames, & McKinstry, 2013; Ritterband et al., 2009; Zachariae, Lyby, Ritterband, & O'Toole, 2016). Four online behavioural parent-implemented interventions for sleep were identified in a recent systematic review of telehealth interventions for sleep problems in children and adolescents (McLay, Sutherland, Machalicek, & Sigafoos, 2020), three of which were for TD infants or children, and one of which was for children with ASD (Roberts et al., 2019). The authors of the review reported that the Roberts et al. (2019) study showed promising treatment effects for sleep in children with ASD, but that more research was required (McLay et al., 2020). To our knowledge, no other online parent-implemented interventions exist for treating sleep in children with NDDs.

Bridging the Gap: Developing an Online Parent-Implemented Intervention for Sleep Problems in Children with NDDs

An online or eHealth parent-implemented intervention may be an ideal way to address the need for an accessible, effective, and evidence-based intervention for sleep problems in children with NDDs, while simultaneously overcoming barriers to treatment provision and access. Evidence suggests that the same strategies that are effective for treating sleep problems in TD children – namely, behavioural intervention – are also effective for children with NDDs, allowing for some modification as needed (Ali et al., 2018; Jan et al., 2008; Malow et al., 2012; Rigney et al., 2018). This is consistent with a growing recognition in both the pediatric and adult sleep literatures that the core components of sleep interventions are transdiagnostic: in other words, similar for the TD population and others such as those with NDDs (Blake, Sheeber, Youssef, Raniti, &

Allen, 2017; Brown et al., 2013; Dolsen, Asnarow, & Harvey, 2014; Harvey, 2009; Harvey, 2016; Harvey et al., 2011; Palermo et al., 2016; Rigney et al., 2018).

Given the growing recognition that the core intervention components are similar for TD and other groups, and the potential utility of an eHealth intervention to address treatment barriers, it seemed appropriate to begin by modifying an existing intervention for pediatric insomnia in TD children. *Better Nights, Better Days* (BNBD) is a parent-implemented eHealth intervention for use with 1- to 10-year-old TD children with insomnia (Corkum et al., 2019; Corkum et al., 2018; Speth et al., 2016; http://betternightsbetterdays.ca). The intervention is based on behavioural strategies that are effective for treating insomnia in TD children, and is a five-session, modular, and self (parent)-directed program that parents can complete at their own pace. Each session is designed to take one to two hours to complete, and involves reading, watching videos, completing activities, and using interactive tools. Intervention content includes: Session 1 – general psychoeducation about children's sleep and sleep problems; Session 2 – healthy sleep practices, bedtime routines, and nap scheduling; Session 3 – strategies for independent settling to sleep at bedtime; Session 4 – strategies for night waking, napping, and early morning awakenings; Session 5 – maintenance and preparing for the future. Parents must complete at least five daily sleep diaries and wait a week to allow time to try implementing each session's strategies before progressing to the next session. Preliminary results of a Canada-wide RCT of BNBD show that the intervention improved children's sleep onset latency and sleep efficiency, as well as children's psychosocial health and quality of life, and parents' fatigue (Corkum et al., 2019; NCT02243501, clinicaltrials.gov).

Four foundational studies, two of which are included in this dissertation, were conducted to build evidence for the development of an intervention for sleep problems in children with NDDs and determine how to adapt BNBD. First, Rigney and colleagues (2018) conducted the aforementioned systematic review of parent-implemented behavioural interventions for pediatric insomnia in NDDs, which established that the same behavioural strategies recommended for TD children can be used in children with NDDs, and across multiple NDD diagnoses. Next, Ali and colleagues (2018) conducted a Delphi study with HCPs who had expertise in sleep and NDDs to identify consensus recommendations for intervention content. Next, I conducted a qualitative exploration of barriers and facilitators to sleep treatment for children with NDDs from both parents' and HCPs' perspectives, to determine what types of barriers to treatment access and implementation such an intervention would need to overcome, as well as identify facilitators to access and implementation. Lastly, in order to determine what modifications the new intervention would require and the extent to which BNBD could be used transdiagnostically for parents of children with NDDs, I conducted a usability study that evaluated whether parents of children with NDDs found the original, unmodified BNBD program to be usable, feasible and acceptable when implemented with their children (Tan-MacNeill et al., 2020). Based on the findings from these four studies, the research team has adapted and modified BNBD into *Better Nights, Better Days for Children with Neurodevelopmental Disorders* (BNBD-NDD), which focuses on parents of children with ASD, ADHD, CP, and FASD (these four diagnoses were included in keeping with the then-focus of the entity that provided funding for the BNBD-NDD project [NeuroDevNet, now Kids Brain Health Network]). A Canada-wide RCT of BNBD-NDD is currently underway (NCT02694003, clinicaltrials.gov).

Research Objectives

This dissertation aimed to contribute to the literature on developing accessible and effective interventions for treating sleep problems in children with NDDs and to the development of BNBD-NDD. The specific research objectives were to: 1) review and synthesize the literature on online parent-implemented interventions for children with NDDs, 2) explore the broader psychosocial context of sleep treatment seeking, access, uptake, and provision for children with NDDs, and 3) evaluate whether a behavioural eHealth parent-mediated intervention for sleep problems in TD children was determined to be usable, feasible, and acceptable by parents of children with NDDs. These goals were achieved via three studies, each contained in a separate manuscript. Chapter 2 consists of a systematic review of online parent-implemented interventions for core symptoms and other associated behaviours in children with NDDs (Study 1). This study does not focus specifically on sleep interventions, but rather provides valuable information related to whether online parent-implemented interventions can be used to deliver EBIs to parents of children with NDDs and is thus connected to Study 3. Chapter 3 is a qualitative exploration of barriers and facilitators to sleep treatment access, implementation, and provision experienced by parents and HCPs of children with NDDs (Study 2). Chapter 4 describes a usability study that used a combination of qualitative and quantitative methods to determine whether the original unmodified BNBD intervention was perceived as usable, feasible, and acceptable by parents of children with NDDs, and what modifications they might suggest to make it more so (Study 3). Chapter 5 concludes this dissertation with a general discussion of the findings of the three studies, clinical implications, and future research directions.

Chapter 2: A Systematic Review of Online Parent-Implemented Interventions for Children with Neurodevelopmental Disorders

The manuscript based on this systematic review is presented below. Readers are advised that Kim M. Tan-MacNeill, under the supervision of Dr. Penny Corkum and Dr. Isabel Smith, was responsible for the research questions, the review methodology, all aspects of the systematic search process, critical analysis of the included studies, and all aspects of the writing process. Critical editorial feedback was provided by dissertation committee members Dr. Shannon Johnson and Dr. Jill Chorney. The following manuscript has been submitted for publication, cited as follows:

Tan-MacNeill, K.M., Smith, I.M., Johnson, S.A., Chorney, J., & Corkum, P.V. (2020). *A systematic review of online parent-implemented interventions for children with neurodevelopmental disorders*. Manuscript submitted for publication.

Abstract

Families of children with neurodevelopmental disorders (NDDs) experience barriers to accessing evidence-based interventions (EBIs). Online parent-implemented interventions can increase treatment access by reducing professionals' direct involvement and allowing parents to implement some forms of treatment at home. The objectives of this systematic review were to: 1) identify online parent-implemented interventions for children with four NDDs – Autism Spectrum Disorder (ASD), Attention-Deficit / Hyperactivity Disorder (ADHD), Cerebral Palsy (CP), and Fetal Alcohol Spectrum Disorder (FASD); 2) assess the quality of studies; 3) assess evidence for efficacy and / or effectiveness; and 4) determine whether any of the interventions met EBI criteria. Five databases were searched. Twelve interventions were identified (ASD = 8, ADHD = 3, CP = 0, FASD = 1). Interventions frequently had modular designs and utilized videos and slideshows; they varied in levels of external support. Various behaviours were targeted (ASD: social communication, imitation, sleep; ADHD: inattention, impulsivity, hyperactivity, externalizing behaviours; and FASD: behavioural regulation). Eight interventions demonstrated improved child outcomes (ASD = 6, ADHD = 2). Despite preliminary evidence that online parent-implemented interventions for children with NDDs are effective, overall study quality was poor and no interventions met EBI criteria; more randomized controlled trials are needed.

Keywords: online, intervention, children, neurodevelopmental disorders, parent-implemented intervention, review

Introduction

Children with neurodevelopmental disorders: Need for evidence-based interventions

Neurodevelopmental disorders (NDDs) emerge early in childhood and are characterized by developmental differences that result in functional impairments across social, personal, academic, and occupational domains (DSM-5, American Psychiatric Association, 2013). The present study focuses on four NDD diagnoses that represent a range of functional impairments: Autism Spectrum Disorder (ASD), Attention-Deficit/Hyperactivity Disorder (ADHD), Cerebral Palsy (CP), and Fetal Alcohol Spectrum Disorder (FASD). Given that children with NDDs experience impairments in many areas, they require intervention for a range of challenges, including core NDD symptoms and associated behaviours. It is critical that families have access to evidence-based interventions (EBIs) to ensure the best outcomes for their children.

Evidence-based interventions refer to interventions supported by empirical evidence of efficacy and/or effectiveness (American Psychological Association, 2006). Various criteria have been developed to operationalize EBIs. For example, according to the APA Division 12 Task Force criteria (Chambless & Hollon, 1998; Chambless & Ollendick, 2001), interventions can be categorized in four ways: well-established, probably efficacious, possibly efficacious, and experimental treatment. Well-established interventions would qualify as EBIs; probably efficacious interventions would be promising; and the other two categories indicate that further research is required. The criteria for these categories are summarized in Table 2.1.

Barriers to accessing evidence-based interventions and potential solutions

Many barriers limit access to EBIs for families of children with NDDs. The first of two key barriers is that families experience logistical and physical barriers (Johnson & DeLeon, 2016). These include financial cost or lack of insurance coverage, living in remote areas, long waits for access to intervention or specialist clinicians, transportation difficulties (e.g., bringing children with functional limitations or behavioural problems to a clinic setting), and parental stress and burden (Almogbel, Goyal, & Sansgiry, 2017; Smith et al., 2014; Thomas, Ellis, McLaurin, Daniels, & Morrissey, 2007; Weisenmuller & Hilton, 2020). A second barrier is that few health care providers are adequately trained to provide EBIs (Weisenmuller & Hilton, 2020; Wright et al., 2015). For example, systemic factors such as limited time and resources can constrain opportunities for health care providers to receive comprehensive training in providing standardized EBIs (Weisenmuller & Hilton, 2020), as well as limit their ability to ensure consistency and continuity of care for families (Wright et al., 2015).

How can such barriers to access be circumvented to make EBIs more available to children with NDDs? Firstly, having parents implement the interventions themselves can help to address these barriers. Parent-implemented interventions are known by various terms, including “parent mediated” and “parent training” interventions. Parent-implemented interventions for NDDs are designed to teach parents to promote behavioural change in their children (e.g., skill acquisition, reduction of NDD symptoms or maladaptive behaviour). They differ from parent support or educational interventions, which benefit children indirectly through promotion of parent knowledge about NDD diagnoses and care (Bearss, Burrell, Stewart, & Scahill, 2015). Parent-implemented interventions focus on children’s outcomes. Parental well-being and self-efficacy are often measured as secondary outcomes, which is important given that they may mediate child intervention outcomes (Sanders & Morawska, 2014; Tarver et al., 2019; Wainer, Hepburn, & McMahon Griffith, 2017). Parent-implemented interventions are effective for a range of behavioural targets in children with ASD (Tarver, Daley, Lockwood, & Sayal, 2014; e.g., language and social communication – Althoff, Dammann, Hope, & Ausderau, 2019) and ADHD (Coates, Taylor, & Sayal, 2015), and are recommended for children with FASD (Paley & O’Connor, 2009) and CP (Whittingham et al., 2011).

Secondly, delivering interventions online (often called eHealth, internet or web-based interventions) can also address barriers to treatment. Internet usage is widespread, with at least 94% of Canadian families having home access to the Internet as of 2019 (Statistics Canada, 2019). The internet may offer specific advantages for the provision of evidence-based interventions, including the ability to ensure that interventions are structured and standardized, interactive and engaging (i.e., including audio, video, and animation content), and customizable to families’ needs and interests, in addition to allowing real-time data collection and monitoring of progress (Rapoff, 2013). Online interventions have been shown to be effective for a range of health, mental health, and behavioural outcomes in both children and adults (Barak & Grohol, 2011; Cushing & Steele, 2011).

Online parent-implemented interventions reduce the need for HCP resources and offer greater accessibility, allowing parents to implement interventions with their children at their convenience and chosen pace from home (Rapoff, 2013). Unlike telehealth or video-based parent-implemented interventions (which involve health care professionals or paraprofessionals teaching parents via telephone or real-time web-/video-conferencing; Lingley-Pottie & McGrath, 2013), online parent-implemented interventions do not require professional involvement. Additionally, within a “stepped care” model of treatment, self-management based treatment is proposed as an initial “step” for psychological care (Richards, 2012). Online parent-implemented interventions may be an ideal and accessible first “step” that reduces burden on the healthcare system.

Emerging evidence for online parent-implemented interventions

Emerging evidence suggests that online parent-implemented interventions can improve outcomes for both children and parents. However, only three small reviews (conducted before 2017) have examined online parent-implemented interventions for specific NDDs. A narrative review of Internet-based interventions for parents of children with ASD and / or intellectual disability included both parent-implemented and parent support interventions, as well as telehealth and videoconferencing (Meadan & Daczewitz, 2015). This review highlighted the potential advantages of online treatment as well as the need for more research, but did not examine the methodological quality of the studies. A review of online ASD resources for parents (including social media, blogs, support groups, and online interventions) found promising evidence of effectiveness, but noted considerable variability in technology used and level of interactivity (Hall, Culler, & Frank-Webb, 2016). A review of remotely delivered parent-implemented interventions for families of children with ASD living outside urban areas counted six online parent-implemented interventions amongst other types of such interventions (Parsons, Cordier, Vaz, & Lee, 2017). They found preliminary evidence that these interventions improved children’s social and communicative behaviour. Other than these ASD-focused reviews, two reviews of parent-implemented interventions included studies with parents of children with ADHD, although ADHD was not the primary focus of either (Baumel, Pawar, Kane & Correll, 2016; Nieuwboer et al., 2013). Both reviews found online parent-implemented programs to be effective in improving child outcomes and changing parenting practices, but noted that heterogeneity amongst interventions (e.g., variations in intensity of treatment and level of support provided) limited generalizability of findings.

Rationale and Research Questions

Existing reviews of online parent-implemented interventions for children with NDDs focus on ASD, do not clearly distinguish between online interventions and other types of digital interventions, and tend to conflate parent-implemented interventions with parent-support interventions. Broadly, the literature calls for more research on online parent-implemented interventions both for typically developing children and those with NDDs (Cushing & Steele, 2011; Hall & Bierman, 2015; Hollis et al., 2017). The present review focuses on online parent-implemented interventions for children with ASD, ADHD, CP, and FASD, carefully excluding telehealth and other types of digital interventions as well as parent-support interventions.

This review aims to address the following four research questions: 1) What is the state of the literature regarding parent-implemented interventions for treating core NDD symptoms and other behaviour in children with ASD, ADHD, CP, and FASD? Specifically, how many parent-implemented online interventions have been studied, what are their general characteristics, and what types of support are provided? 2) What is the quality of the studies that have evaluated these interventions? 3) What is the evidence for the efficacy and / or effectiveness of these interventions? 4) Do any current online parent-implemented evidence-based interventions meet EBI criteria?

Method

Information sources and search strategy

Published studies were identified through searching four electronic databases: PsycINFO (EBSCOHost), CINAHL (EBSCOHost), PubMed, and Scopus. A systematic search of unpublished dissertations and theses was conducted using Proquest Dissertations and Theses. The search was constructed to include four key semantic concepts, using the Boolean operator 'OR' to concatenate terms within each concept, and the Boolean operator 'AND' to concatenate the four concepts. The first term identified the broad target population (children), the second term further specified the target population (with NDDs), the third term identified interventions, and the fourth term further specified the online or web-based aspect of the intervention. The terms were:

1. Children (e.g., child, adolescent, toddler, schoolchildren, minor, juvenile, paediatric)
2. Neurodevelopmental Disorders (e.g., ASD, ADHD, CP, FASD, and closely related NDDs / broad categories such as communication disorder, social communication disorder, intellectual disability, developmental delay)
3. Intervention (e.g., treatment, therapy, program)
4. Online (e.g., eHealth, web-based, internet-based)

Specific search terms and concepts and a sample search strategy are available in Appendix A. The search strategy specified title / abstract / keyword / subject and was tailored to each database, with terms mapped to either Medical Subject Headings (MeSH) or database-specific subject headings (e.g., PsycINFO's Thesaurus). Searches included articles available up to January 1, 2020. Additional studies were identified through pearling (hand-searching the reference lists of included studies) and searching the archives of the *Journal of Medical Internet Research* and *Internet Interventions*. Figure 2.1 depicts the search process as a Preferred Reporting Items for Systematic Reviews (PRISMA; Moher, Liberati, Tetzlaff, Altman, & The PRISMA Group, 2009) diagram.

Eligibility criteria

Studies were included if they met these criteria:

1. Population/Participants: Parents of children with NDDs aged 0-18 years. Target NDDs included: ASD (also Pervasive Developmental Disorder), ADHD (also Attention Deficit Disorder [ADD]), CP, FASD (also Fetal Alcohol Effects [FAE], Prenatal Alcohol Exposure [PAE], Alcohol-Related Neurodevelopmental Disorder).
2. Intervention Type:
    i. Online: Intervention content was delivered through a website or online platform.
    ii. Parent-implemented intervention: Interventions taught parents strategies and explicitly required parents to implement these strategies with their children. Interventions were thus designed to benefit children directly.

Interventions had to address core symptoms of NDDs and other associated behaviour. Parent-support interventions (e.g., psychoeducational interventions designed to benefit children indirectly) were excluded. Interventions could be standalone/fully self-directed or include external support (e.g., weekly phone calls with a therapist to answer questions; online feedback; technical support). Telehealth and distance-delivered therapy through real-time video-conferencing or phone were excluded.

3. Study Design: Studies were tests of efficacy or effectiveness (e.g., single-case experimental designs, pre-/post- study designs, randomized controlled trials [RCTs]). Multiple articles pertaining to the same study were grouped.

Study selection

Initial search results from each database were combined, then duplicates were removed first using RefWorks reference manager software and then using Covidence systematic review management software. In Covidence, results were screened by titles/abstracts to determine whether they broadly met inclusion criteria, followed by full-text screening.

Data extraction

Following title, abstract, and full-text screening, included studies were fully reviewed and information was extracted. Data extraction was organized using the PICOS framework to identify populations/problems, interventions, comparison, outcomes, and types of studies (Schardt, Adams, Owens, Keitz, & Fontelo, 2007).

Quality assessment

The methodological quality of each study was evaluated and scored according to the quality index for randomized and non-randomized studies proposed by Downs and Black (1998). This is a 27-item checklist that includes five sub-scales (1. reporting, 2. external validity, 3. internal validity – bias, 4. internal validity – confounding, 5. power). Numerous studies in this review did not report sufficient data to calculate power, so this subscale was excluded. As such, this review utilized 26 items, allowing for a maximum score of 27 points (each item is scored 0 or 1, except for one item which is scored 0 to 2; see Table 2.2 for the checklist). Higher scores indicate higher quality. Previous reviews have used this modified quality index (e.g., Rigney et al., 2018). The Downs and Black (1998) quality index has demonstrated good test-retest reliability ($r = 0.88$), high correlations with other validated quality assessment instruments used for non-randomized studies ($r = 0.86 - 0.90$), and high internal consistency (Kuder-Richardson 20: 0.89). Following the quality assessment, interventions were classified into one of four categories for evidence-based interventions (well-established, probably efficacious, possibly efficacious, or experimental), according to the aforementioned criteria established by the APA Division 12 Task Force and detailed in Table 2.1.

Results

Results are organized as follows: search results; descriptions of the interventions (content, target outcomes, delivery characteristics); descriptions of the studies (study design, participants, and comparison groups); quality assessment ratings of studies; and evaluation of evidence for efficacy / effectiveness of the interventions.

Search Results

See Figure 2.1 for the PRISMA flow diagram. The search identified a total of 6431 references, which was reduced to 4170 after removing duplicates. An additional 3924 references were removed after title and abstract reviews. Of the 246 references reviewed in full-text, 231 were excluded and 15 remained. The primary reasons for exclusion were that interventions were telehealth-based (i.e., provided therapy via videoconferencing or telephone; $n = 63$) or not parent-implemented (i.e., were self-implemented by child, therapist-implemented, or school-based; $n = 56$). An additional two peer-reviewed articles were identified through pearling (based on included studies' reference lists). Therefore, 17 references met the inclusion criteria. Three references pertained to the same study. Overall, there were 15 studies that evaluated 12 distinct interventions. Eight interventions focused on children with ASD, three on ADHD, and one on FASD. No studies were found for children with CP. Table 2.3 summarizes intervention characteristics (target, design, level of support). Table 2.4 contains a summary of the research studies evaluating these interventions and the quality assessment ratings.

Description of Interventions

Interventions by NDD Group

Autism Spectrum Disorder. Eight parent-implemented interventions for children with ASD were identified across nine studies.

Three interventions focused on improving social communication skills, a key area of impairment in ASD. The ImPACT Online intervention, developed by Ingersoll and colleagues (Ingersoll, Wainer, Berger, Pickard, & Bonter, 2016; additional study information in Ingersoll & Berger, 2015; Pickard, Wainer, Bailey, & Ingersoll, 2016), is a naturalistic, developmental behavioural intervention (NDBI – Schreibman et al., 2015) that aimed to improve children's spoken language during parent-child interactions. Another intervention, the Pivotal Response Treatment (PRT) online course, promoted social communication skills such as vocalizations, eye contact, and positive affect in children with ASD (McGarry, Vernon, & Baktha, 2019). The POWR online communication partner training model aimed to teach parents of children with ASD and complex communication needs (i.e., limited verbal ability) how to increase their children's communication by providing more communication opportunities (Douglas, Kammes, & Nordquist, 2018).

Improvements in children's imitation skills, another core area of impairment in ASD, were targeted by the online Reciprocal Imitation Training (RIT) intervention (Wainer & Ingersoll, 2013, 2015). Another intervention, the Enhancing Interactions tutorial, was aimed at improving children's engagement and participation in daily home routines (e.g., bath time, play time; Ibanez, Kobak, Swanson, Wallace, Warren, & Stone, 2018).The Applied Behaviour Analysis (ABA) Web-Based Training Program was designed to teach parents to use ABA strategies to manage children's problem behaviour, increase communication skills, and improve parent-child interactions (Blackman, Jimenez-Gomez, & Schvarts, 2019). A Facebook-delivered intervention called Project CHASE (Children with Autism Supported to Exercise) aimed to teach parents to increase children’s physical activity (Healy & Marchand, 2019). Finally, another study examined an online Parent Sleep Education intervention, which taught parents to implement behavioural strategies to improve children’s sleep (Roberts, Smith, & Sherman, 2019).

Attention-Deficit / Hyperactivity Disorder. Three parent-implemented interventions for core symptoms of ADHD were identified across five studies. All three interventions were based on behavioural parent training (BPT), a type of behaviour management intervention that teaches parents to change negative parenting practices to more positive practices in order to reduce or eliminate ADHD symptoms (e.g., inattention, hyperactivity) and externalizing behaviour, as well as improve interactions between parents and children.

The first BPT program, Promoting Engagement for ADHD pre-Kindergartners (PEAK), taught parents how to modify their children’s behaviour at home (DuPaul, Kern, Belk, Custer, Daffner, Hatfield, & Peek, 2018a; DuPaul, Kern, Belk, Custer, Hatfield, Daffner, & Peek, 2018b – Step 5 sample). The next BPT program was designed to teach parenting skills to reduce children’s externalizing behaviour (Breider, de Bildt, Nauta, Hoekstra, & van den Hoffdakker, 2019). This program was referred to as “blended” because online training was supplemented by supportive therapist contact. Thirdly, the Triple P Online (TPOL) positive parenting program was examined in samples of preschoolers with ADHD symptoms (Franke, Keown, & Sanders, 2017; Day & Sanders, 2018). This online intervention is an adaptation of Level 4 of the Triple P Positive Parenting Program (a multilevel system of behavioural parent training interventions aimed at reducing or preventing social, behavioural, and emotional problems in preschoolers), which has shown efficacy in improving problematic child behaviours in children with early-onset conduct problems (Sanders, Baker, & Turner, 2012).

Fetal Alcohol Spectrum Disorder. An online program that taught parents how to teach their children positive behavioural regulation skills and provided information about FASD and FASD advocacy was adapted from an in-person group workshop, with the aim of improving children's behavioural regulation and internalizing and externalizing behaviour, as well as parent knowledge (Kable, Coles, Strickland, & Taddeo, 2012).

Intervention Content and Delivery Characteristics

Most interventions were modular *(n* = 9), consisting of short (< 1 hour), sequential modules or sessions designed to be completed in a specific time period (e.g., one per week). The number of modules or sessions ranged from three to 12. Programs ranged in duration from 1 to 12 weeks. In two interventions, progression to the next module was contingent on parents completing a comprehension quiz (Breider et al., 2019) or submitting a video of themselves demonstrating correct strategy use (McGarry et al., 2019).

Non-modular intervention designs (*n* = 3) included audiovisual podcasts with online blogging (Roberts et al., 2019), a web page containing a narrated PowerPoint presentation (Kable et al., 2012), and a private Facebook group (Healy & Marchand, 2019). Most interventions contained text, narrated slideshows / PowerPoint presentations (*n* = 8), and video modelling of strategies (*n* = 8). Several interventions included downloadable written materials such as manuals or summary sheets (*n* = 4), online quizzes (*n* = 5), or online resource pages (*n* = 1). Two interventions were customizable (e.g., choosing which behaviours to treat – Ibanez et al., 2018; generating individualized progress workbooks – Franke et al., 2017; Day & Sanders, 2018).

Intervention Level of Support

The level of external support provided to parents varied across interventions, ranging from technical support only to face-to-face supportive meetings. “Self-directed” refers to parents’ use of the intervention without any form of external support. Of note, five studies compared self-directed use of the interventions with therapist-assisted use of the interventions, either in separate treatment conditions or through a multiple baseline design; external support within therapist-assisted treatment conditions is described in the “Study Comparisons” section below (p. 42). This section only describes external support associated with the original form of the online intervention.

Six interventions were self-directed, only allowing technical website or software support (POWR Online Communication Training – Douglas et al., 2018; PRT online training – McGarry et al., 2019; RIT – Wainer & Ingersoll 2013, 2015; ABA Web-Based Training – Blackman et al., 2019; Enhancing Interactions – Ibanez et al., 2018; FASD Education and Training – Kable et al., 2012). Additionally, one of the two studies evaluating the TPOL intervention examined a fully self-directed version with no assistance except technical support upon request (Day & Sanders, 2018), whereas the second examined TPOL with external support (Franke et al., 2017; described below). In the Project CHASE Facebook-delivered intervention (Healy & Marchand, 2019), parents’ engagement with the Facebook group was self-directed, but the researchers posted psychoeducational information and sent motivational reminders to participants once per week via Facebook Messenger. In the Parent Sleep Education intervention, parents were able to email questions to the instructor, and the instructor also posted discussion questions for parents to respond to in their blogs.

More substantive forms of external support were noted in the three ADHD-focused interventions, including therapist assistance or supportive coaching. In the PEAK intervention (DuPaul et al., 2018a, 2018b), parents received weekly phone calls from a consultant or research assistant to review the strategies from the online sessions and answer questions about implementation. For the Blended BPT intervention (Breider et al., 2019), the main content of the intervention was reportedly provided to parents through the six online modules. However, training began and ended with a clinic-based face-to-face contact with a therapist, including an initial 90-minute meeting to introduce parents to the program and select target behaviours and situations. Parents also received four 45- to 60-minute evaluation contacts focused on their progress throughout the intervention. Therapists were instructed to give parents online feedback on each exercise, to provide parents with access to the next module contingent on understanding of previous content, to remind parents to log in if there was no online activity for two weeks or more, and to schedule an additional face-to-face contact if they felt that parents were experiencing difficulty with the intervention. Finally, the level of support in the two studies examining TPOL online differed, as noted above. In the study by Franke and colleagues (2017), parents received two phone consultations with an intervention facilitator to tailor strategies to their needs and to solve problems.

Description of Studies

Study Design

Three studies were RCTs, and four were pilot RCTs with smaller samples. Sample sizes of RCTs ranged from 22 to 183 participants. The remainder were pre- /post-test designs (*n* = 5) and single-case, multiple baseline / probe designs (*n* = 3). For pre- /post-test studies, sample sizes ranged between 8 and 25. Single-case studies included 3 to 5 parent-child dyads.

Study Participants

Interventions were aimed at parents of children whose ages ranged from 20 months (1.7 years) to 16 years. Most studies (*n* = 10) did not report information on comorbid conditions, but these were common within the studies that did (*n* = 5). For studies focusing on ASD, comorbid conditions included ADHD, anxiety, speech delay / apraxia, sensory processing disorder, learning disability, intellectual disability, and epilepsy (Douglas et al., 2018; Healy & Marchand, 2019; Roberts et al., 2019). For studies focusing on ADHD, comorbidities included oppositional defiant disorder, disruptive behaviour disorder, Tourette syndrome, and anxiety (Breider et al., 2019; DuPaul et al., 2018a).

Across studies, most participants were English-speaking mothers, married or living with a partner, and well-educated, with most parents having at least some college / university education. Only six studies reported mean parental ages (Day & Sanders, 2018; Douglas et al., 2018; Franke et al., 2017; Healy & Marchand, 2019; Ibanez et al., 2018; Roberts et al., 2019), which ranged between 34 and 41 years. Socio-demographic characteristics differed considerably across studies. One study recruited from underserved treatment areas (Ingersoll et al., 2016), two recruited participants from remote locations (Healy & Marchand, 2019; Wainer & Ingersoll, 2015), and one sought participants with greater socioeconomic / family risk factors (Day & Sanders, 2018). Four studies explicitly excluded parents who had experience with the type of therapy the study was evaluating (Blackman et al., 2019; Breider et al., 2019; Wainer & Ingersoll, 2013, 2015).

Study Comparisons – Groups or Intervention Phases

Twelve studies included a comparison group, among which six studies included a waitlist control (WLC) group; of these studies, two only included a WLC or delayed intervention comparison group (Ibanez et al., 2018; Franke et al., 2017). Two studies compared intervention use with and without therapist assistance in the form of telephone coaching once per week (Day & Sanders, 2018) or remote coaching twice per week (Ingersoll et al., 2016) Three studies used a multiple baseline or multiple probe design to compare initial self-directed intervention phases with subsequent paraprofessional- or therapist-coached intervention (Douglas et al., 2018; Wainer & Ingersoll, 2013, 2015). Five studies compared online and face-to-face delivery of the same intervention content (Blackman et al., 2019; Breider et al.; 2019; DuPaul et al., 2018a; Kable et al., 2012; Roberts et al., 2019).

Quality Ratings for Studies Evaluating the Interventions

The average total score for methodological quality (Downs & Black, 1998) was 17.1 (SD = 2.5, 65.9%), ranging from 12 to 21 (46.2 – 80.8%) out of 27 points. The eight interventions for children with ASD had a mean quality rating of 16.7 (SD = 1.9, 61.7%). The three interventions for children with ADHD had a mean quality rating of 17.8 (SD = 3.7, 65.9%). The quality rating for the single FASD intervention was 18 (66.7%). Across all studies, the mean quality rating for RCT studies was 19.1 (SD = 1.6, 70.9%), whereas the mean quality rating for pre/post studies was 15.4 (SD = 2.3, 57%) and for single-case studies, 15.3 (SD = 0.6, 56.8%).

Most studies received high scores on the *reporting subscale* (although no studies reported on adverse events). All studies scored poorly on the *external validity subscale*: no studies adequately demonstrated that their samples were representative of the population or that participants were representative of the recruitment sample. Only two studies utilized a treatment / intervention that was representative of interventions already in use in the source population (TPOL – Day & Sanders, 2018; Franke et al., 2017). Studies scored higher on the *internal validity – bias subscale*, indicating that most used appropriate statistical tests, used reliable outcome measures, and demonstrated that participants had reliable compliance with interventions. However, only one study blinded participants to treatment condition (Ingersoll et al., 2016), and no studies attempted to blind measurement of the main outcomes. Most studies also scored poorly on the *internal validity – confounding subscale*. This subscale was affected by whether participants were recruited during a comparable time period, were randomized (only seven studies did so), whether randomization was concealed, and whether analyses were adequately adjusted for confounders.

Efficacy and/or Effectiveness Data

Eight of the twelve interventions showed clear evidence of efficacy or effectiveness for improving targeted child outcomes (i.e., statistically significant results), and one intervention showed some mixed evidence of improved outcomes. Three interventions did not significantly improve child outcomes. Of the nine interventions that examined parent outcomes (including parent knowledge, stress, self-efficacy, and mental health), all reported improvements in at least some of these outcomes. Below, outcome results for children and parents are reviewed for each intervention by NDD group, followed by a summary of findings regarding treatment comparison groups.

Autism Spectrum Disorder

Child Outcomes. All three of the social communication interventions improved children's communication behaviours and language targets (Douglas et al., 2018; Ingersoll et al., 2016; McGarry et al., 2019). The POWR Online Communication Partner Training program was evaluated by a single-case, multiple probe design study and found to increase both parent-provided communication opportunities and children's communication skills (Douglas et al., 2016). ImPACT Online was tested in a pilot RCT study and found to improve children's language, but social skills only improved in the therapist-assisted condition (see section 3.5.4, below; Ingersoll et al., 2016). A full-scale efficacy trial of ImPACT Online is currently underway (NCT02721381, clinicaltrials.gov). The PRT online course, which was evaluated in a small pre/post study, improved social communication behaviours for children (McGarry et al., 2019).

The Enhancing Interactions intervention was evaluated in an RCT and found to increase parent-reported child engagement behaviours during home-based routines

(Ibanez et al., 2018). Use of the ABA Web-Based Training Program, evaluated in a pre/post study, resulted in significantly improved and more positive parent-child interactions (Blackman et al., 2019). The Parent Sleep Education intervention significantly increased total sleep time and decreased night waking for children, as measured by both parent report and actigraphy in a pre/post study (Roberts et al., 2019).

Several studies showed mixed results or had non-statistically significant but promising results. Findings for the RIT imitation intervention, which was evaluated in two single-case multiple baseline studies, were mixed (Wainer & Ingersoll, 2013, 2015). Wainer and Ingersoll (2013) found substantial increases in imitation rates for two out of three children during the self-directed phase of treatment, whereas Wainer and Ingersoll (2015) only found small increases in imitation (at differing rates) for four out of five children during the self-directed phase. Imitation rates continued to increase in the subsequent coached phases of treatment for both studies, leading to inconclusive results regarding overall effectiveness of self-directed RIT. In the small pre/post study examining Project CHASE (Healy & Marchand, 2019), activity increased for all child participants, but the change in parent-reported physical activity was not statistically significant due to lack of power.

Parent Outcomes. Six of the ASD-focused interventions also examined parent outcomes, including self-efficacy, stress, and knowledge. Two interventions, ImPACT Online (Ingersoll et al., 2016) and Enhancing Interactions (Ibanez et al., 2018), improved parent self-efficacy and lowered parent stress. ImPACT Online also increased parents' positive perceptions of their children (Ingersoll et al., 2016). The Parent Sleep Education intervention significantly improved parents' quality of life and decreased their fatigue

(Roberts et al., 2019). Two interventions significantly increased parents' knowledge of specific content – the RIT intervention increased knowledge of imitation in both studies (Wainer & Ingersoll, 2013, 2015), and the ABA Web-Based Training program increased knowledge of applied behaviour analysis (Blackman et al., 2019).

Attention-Deficit / Hyperactivity Disorder

Child Outcomes. Triple P Online significantly improved parent-rated ADHD symptoms and children's social functioning and teacher-rated prosocial behaviour in two RCT studies (Day & Sanders, 2018; Franke et al., 2017). Also in an RCT, PEAK was found to significantly reduce parent-reported child ADHD symptoms and improve parent-reported child mood/affect (DuPaul et al., 2018a). The Blended BPT program (Breider et al., 2019) did not significantly reduce the severity of children's externalizing behaviour problems. However, the online intervention group suffered from extremely high attrition (90% drop-out by the last intervention module).

Parent Outcomes. No parent outcomes were measured in the Blended BPT study (Breider et al., 2019), but the studies evaluating TPOL and PEAK examined a range of parent outcomes. Both studies found that TPOL significantly improved parenting practices (e.g., reductions in parenting over-reactivity) and significantly reduced parent stress, depression, and anxiety (Day & Sanders, 2018; Franke et al., 2017). Day and Sanders (2018) found that the intervention improved parenting confidence and reduced parental anger at five-month follow-up. Franke and colleagues (2017) found that the intervention significantly increased self-reported parenting competence. In the PEAK

RCT, DuPaul and colleagues (2018a) found that the intervention significantly improved parents' knowledge, but did not significantly reduce parental stress.

Fetal Alcohol Spectrum Disorder

Child Outcomes. The online version of FASD Education and Training for Behavioural Regulation was evaluated in an RCT that did not find significant improvements in parent-reported child behaviour.

Parent Outcomes. The intervention significantly improved parents' knowledge about FASD and FASD advocacy and behavioural management skills.

Comparisons with Waitlist Control Groups, therapist assistance, and face-to-face delivery

The six studies that included a WLC group all found improved child outcomes for the intervention groups relative to controls (ASD: Ibanez et al., 2018; Franke et al., 2017; ADHD: Day & Sanders, 2018; Franke et al., 2017; DuPaul et al., 2018a; FASD: Kable et al., 2012). Studies that compared intervention use with and without therapist assistance found greater improvements with therapist assistance. For example, when parents of children with ADHD received telephone assistance with the TPOL intervention, greater decreases were seen in intensity of difficult child behaviours, as well as negative parenting styles that directly affected children's behaviour (Day & Sanders, 2018), although both conditions (with and without therapist assistance) showed better outcomes than WLC. Similarly, therapist assistance in ImPACT Online resulted in improved social skills for children with ASD compared to the group that did not receive such assistance

(Ingersoll et al., 2016). Additionally, coaching (a form of therapist assistance) enhanced children's outcomes in the ASD-focused intervention studies that utilized a multiple baseline design (Douglas et al., 2018; Wainer & Ingersoll, 2013, 2015).

Findings were mixed regarding the efficacy / effectiveness of online versus face-to-face delivery of intervention content. Nearly all studies found that *both* types of delivery improved child and/or parent outcomes, except for the online blended BPT intervention for ADHD (Breider et al., 2019). However, given the online group's high attrition, it is difficult to draw any conclusions. Two studies found no significant differences between types of delivery (Blackman et al., 2019; DuPaul et al., 2018b). Roberts et al. (2019) found that only online delivery of their psychoeducational sleep intervention resulted in improved / sustained reduction in parent fatigue and decreased night waking for children. In contrast, Kable et al. (2012) found that only face-to-face group delivery of their intervention for core symptoms of FASD produced changes in children's behavioural outcomes, whereas parent outcomes were improved by both face-to-face and online delivery.

Discussion

The purpose of this systematic review was to address four research questions – examining the state of the literature on online parent-implemented interventions for children with NDDs, determining the quality of studies that evaluated these interventions, assessing evidence of efficacy and/or effectiveness, and finally determining whether any of the interventions qualified as EBIs. Overall, the findings show that an increasing number of online parent-implemented interventions have been developed. The overall quality of studies evaluating of these interventions varies considerably: few interventions have been adequately tested. Although evidence of efficacy and/or effectiveness is promising for core symptoms and other behaviour in children with ASD and ADHD, the single FASD intervention was not effective, and no interventions were identified for children with CP. More research is required as currently no online parent-implemented interventions can be considered EBIs.

Current State of the Literature on Online Parent-Implemented Interventions for Children with NDDs

Twelve online parent-implemented interventions for treating core NDD symptoms and other behaviours in children with ASD, ADHD, and FASD were identified, but no interventions for children with CP were found. All were behavioural interventions, requiring parents to implement strategies directly with children, or to change their own parenting practices in order to influence children's behaviour. Online interventions that incorporate behavioural methods have been shown to produce larger effect sizes for target outcomes (Cushing & Steele, 2010). Eight interventions were for ASD, targeting communication skills, imitation, child engagement in daily home routines, parent-child interactions, physical activity, and sleep. Three interventions were for ADHD and based on behavioural parent training (BPT), with the aims of reducing ADHD symptoms and externalizing behaviours and improving parenting practices. One FASD intervention for improving children's behavioural regulation was identified.

Although some common design features were noted across interventions, including modular designs and content delivery via videos and slideshows, the variety of design and support features precludes drawing any conclusions about what features are associated with effectiveness. For example, the level of external support provided to parents varied. Six interventions were fully self-directed, allowing only technical support. The three ADHD-focused interventions each had more substantive external support, including weekly phone consultations or remote coaching, or face-to-face progress and evaluation meetings.

Quality of Studies

The overall quality of studies was weak according to Downs and Black criteria (1998), with an average rating of 65.9%. Only seven studies utilized RCT designs (four of which were pilot RCTs). The small sample sizes may reflect the emerging state of research on online parent-implemented interventions, and make it difficult to draw firm conclusions about efficacy and effectiveness. The methodological quality of studies included in this review also varied considerably. For example, few studies included WLC or treatment comparison groups; some suffered from high attrition; and few provided detailed information about parent participants (including familiarity with and use of the internet). Problematically, child outcomes were frequently evaluated using parent-report measures (e.g., checklists of behaviours) rather than objective assessment or observation, which is concerning given that parents were implementing the treatments. The quality of the descriptions of the interventions varied across studies, with some providing inadequate information on how content was delivered online. Complicating comparisons across interventions, both content delivery and intervention design were heterogeneous (e.g., modular sessions vs. standalone webpage; straightforward adaptations of face-to-face interventions vs. modular interventions designed specifically for online delivery).

Evidence for Efficacy and Effectiveness

Eight of the twelve identified interventions significantly improved child outcomes, including six ASD interventions that improved children's social communication skills (Douglas et al., 2019; Ingersoll et al., 2016; McGarry et al., 2019), parent-child interactions (Blackman et al., 2019), engagement in daily routines (Ibanez et al., 2017), and sleep (Roberts et al., 2019), and two ADHD interventions that improved child ADHD symptoms and externalizing behaviours (Day & Sanders, 2018; DuPaul et al., 2018a, 2018b; Franke et al., 2017). Findings for an intervention to improve imitation skills in children with ASD were mixed (Wainer & Ingersoll, 2013, 2015). While an intervention to promote physical activity in children with ASD showed a trend towards effectiveness, results were not significant (Healy & Marchand, 2019). The single FASD intervention to improve behavioural regulation was not effective (Kable et al., 2012). Most interventions also led to improvements in secondary parent outcomes such as parent stress, knowledge, and parenting self-efficacy, which is important given that parenting behaviour and parents' well-being may mediate the effects of parent-implemented interventions (Sanders & Morawska, 2014).

Several studies found that face-to-face delivery of the same intervention content resulted in greater improvements in child and parent outcomes, compared to online delivery. Parents who took part in face-to-face intervention training may have benefited from the ability to ask questions, or from interacting with other participants within group workshops. The presence of a therapist may also have created more accountability and motivation, increasing parent adherence and treatment fidelity. Similarly, studies that examined therapist-assisted intervention as a comparison group found that it was associated with greater improvements in both children's and parents' outcomes (e.g., Ingersoll et al., 2016; Wainer & Ingersoll, 2015; Day & Sanders, 2018). This finding is consistent with previous reviews of online (Nieuwboer et al., 2013) and face-to-face parent-implemented interventions (O'Brien & Daley, 2011; Tarver et al., 2014), which found that therapist support can enhance outcomes. However, one study found greater improvements in the self-directed online group than the face-to-face group (Roberts et al., 2019), suggesting than online interventions can be as effective or more effective than other formats. Thus, more research comparing interventions with and without therapist assistance / external support is required to determine whether online parent-implemented interventions require support to be effective, or the conditions under which support might be required.

Regarding intervention design, all but one of the modular interventions were associated with improved child outcomes. The single modular intervention that was not effective (Breider et al., 2019) condensed the content of 16 in-person sessions into six multi-part modules. This may have influenced effectiveness and the study was further compromised by high attrition. Neither the standalone web-page design intervention (Kable et al., 2012) nor the private Facebook group intervention (Healy & Marchand, 2019) was found to be effective. Five interventions were adapted from interventions with prior evidence of efficacy or effectiveness. Triple P Online had demonstrated effectiveness with parents of children with conduct problems (Sanders et al., 2012), POWR online communication training was effective when implemented by para-educators (Douglas et al., 2014), and face-to-face versions of ImPACT, RIT, and PEAK had demonstrated effectiveness (respectively: Ingersoll & Dvortcsak, 2010; Ingersoll & Gergans, 2007; DuPaul & Kern, 2011; Kern et al., 2007). Each of the adapted versions of these interventions (i.e., those included in the review) found some evidence of improved child and parent outcomes. Further research is required to determine how or whether these aspects of intervention design are associated with effectiveness.

Qualification as EBIs

None of the interventions identified in this review qualify as well-established interventions for improving core NDD symptoms and other associated behaviours based on the criteria of APA Division 12 Task Force criteria (Chambless & Hollon, 1998; Chambless & Ollendick, 2001; see Table 2.1). Five of the ASD-focused interventions meet criteria to be classified as possibly efficacious, including POWR Online Communication Training (Douglas et al., 2019), ImPACT Online (Ingersoll et al., 2016), PRT Online Training (McGarry et al., 2019), Enhancing Interactions (Ibanez et al., 2017), and Parent Sleep Education (Roberts et al., 2019). Three interventions can be considered experimental treatments, given their mixed results or lack of effectiveness for improving child outcomes: ABA Web-Based Training (Blackman et al., 2019), online RIT (Wainer & Ingersoll, 2013, 2015), and Project CHASE (Healy & Marchand, 2019). Of the ADHD-focused interventions, TPOL may be categorized as probably efficacious (Day & Sanders, 2018; Franke et al., 2017); PEAK can be categorized as possibly efficacious and requiring further study (DuPaul et al., 2018a, 2018b); and Blended BPT online training constitutes an experimental treatment at this time (Breider et al., 2019). Finally, the single FASD Education and Training intervention can be categorized as a possibly efficacious treatment, requiring further evaluation.

Implications and Research Directions

Online parent-implemented interventions could potentially increase families' access to much-needed treatment to address a wide variety of behavioural concerns in children with NDDs and concurrently reduce the burden on health care professionals. However, in spite of emerging evidence that suggests online parent-implemented interventions hold promise, none of the existing interventions can currently be classified as well-established EBIs. More research is needed – namely, methodologically rigorous full-scale RCTs with WLC and alternative treatment comparison groups, well-characterized samples, detailed descriptions of intervention content and delivery, and objective measures of outcomes. Future studies should also examine the impact of external supports, such as therapist assistance, on intervention effectiveness and parents' utilization and adherence. The lack of studies examining online parent-implemented interventions for parents of children with CP and FASD suggests that research with these populations is sorely needed, as well as research on interventions for children with other NDD diagnoses not included in this review. In addition to the development of effective interventions, the field would benefit from cost-effectiveness research that examines trade-offs between magnitudes of differences in outcomes and resources needed (e.g., therapist assistance). Finally, future intervention studies need to distinguish clearly between parent-implemented and parent support interventions. Inconsistent use of terminology muddies the parent-implemented intervention literature (Bearss et al., 2015).

Fortunately, published RCT protocols and usability studies indicate that considerable research on development and evaluation of online parent-implemented interventions for children with NDDs is under way. Interventions in development include the Strongest Families parent training intervention for children with FASD (Hundert et al., 2016; Turner et al., 2015), the Better Nights, Better Days for Children with Neurodevelopmental Disorders eHealth behavioural sleep intervention for children with ASD, ADHD, CP, and FASD (Tan-MacNeill et al., 2020), an interactive behavioural skills training program to reduce challenging behaviour in children with ASD and intellectual disability (Marleau, Lanovaz, Gendron, Higbee, & Morin, 2019), and the aforementioned registered ImPACT Online trial (NCT02721381, clinicaltrials.gov).

Limitations

The present review included a small number of studies. Due to the heterogeneity of the studies identified (e.g., outcomes measured, study populations), meta-analytic evaluation was not possible. The use of inconsistent language to describe interventions implemented by parents in the broader literature necessitated that the present review not only utilize the term “parent-implemented intervention,” but also that search results be carefully examined so as not to conflate different types of parent interventions. Furthermore, although this review focuses on four specific NDDs, interventions directed toward children with other NDDs should be considered for future reviews. Finally, in focusing on efficacy / effectiveness studies with children’s behaviour as the primary outcome, this review did not include studies of other online parent-implemented interventions for children with NDDs that are currently being tested for usability, feasibility, and acceptability, nor did it include pilot studies that only examined changes in parent outcomes such as knowledge.

Conclusion

To our knowledge, this is the first systematic review to examine online parent-implemented interventions for core NDD symptoms and other behaviour in children with a range of NDD diagnoses. Although findings suggest that most published interventions show promising evidence of effectiveness for a variety of behaviours in children with ADHD and ASD, no interventions can currently be classified as EBIs. The overall methodological quality of the identified studies indicates that more research is needed about this potential method of overcoming barriers to intervention access for families of children with NDDs.

Table 2.1

APA Division 12 Task Force Criteria for Evidence-Based Interventions

Category	Criteria
1. Well-established	Demonstrate efficacy in at least 2 between-group designs or a large series of single-case designs (*n* ≥ 9). Studies must be methodologically sound and well-characterized, utilize treatment manuals/clearly describe intervention content. Efficacy must be demonstrated by at least two different investigators.
2. Probably efficacious	Demonstrate efficacy in: a) at least two studies that show superiority to waitlist control, b) at least one between-group design study by the same investigator, or c) a small series of single case design studies (*n* ≤ 3). Studies must be methodologically sound and well-characterized, utilize treatment manuals/clearly describe intervention content.
3. Possibly efficacious	Benefits at least three participants in methodologically sound research by a single investigative team with no conflicting evidence. Requires further research.
4. Experimental	Intervention is evaluated in studies that do not meet criteria for any of the other three categories. Requires further research.

Table 2.2

Modified Downs and Black (1998) checklist used to determine methodological quality of studies included in review

Subscale	Item #	Specific Criteria
Reporting	1	Clear description of study purpose (hypothesis / objective(s))
	2	Main outcomes defined in Introduction or Methods sections
	3	Clear description of participant characteristics
	4	Clear description of intervention of interest
	5	Description / list of principal confounders provided
	6	Clear description of main study findings
	7	Information about random data variability for main outcomes
	8	Reported important adverse events that may be consequence of intervention
	9	Described characteristics of participants lost to follow-up
	10	Reported actual probability values for main outcomes
External validity	11	Sample representative of entire population from which it was recruited
	12	Actual participants included in sample representative of entire population
	13	Intervention representative of that in use in source population
Internal validity - bias	14	Attempt to blind study participants
	15	Attempt to blind those measuring main intervention outcomes
	16	Clear description of results not based on a priori hypotheses (data dredging)
	17	Analyses adjusted for different lengths of follow-up
	18	Appropriate statistical tests used to assess main outcomes
	19	Reliable compliance with the intervention
	20	Accurate, valid, and reliable main outcome measures
Internal validity – confounding (selection bias)	21	Comparability of individuals included in all comparison groups in relation to the population recruited from
	22	Comparability of individuals included in all comparison groups in relation to the period of time during which they were recruited
	23	Randomization of participants to study group
	24	Randomization concealed from participants / those conducting intervention
	25	Adequate adjustment for confounders in the analyses from which main findings were drawn
	26	Losses to follow-up taken into account for analyses

Note: all items scored 0 or 1, except item 5 (scored 0, 1 or 2).

Table 2.3

Summary of online parent-implemented intervention characteristics (target, design, and level of support).

Intervention, by NDD	Target of Intervention	# Sessions / Duration	Online Content Delivery	Level of Support
ASD				
POWR – Online Communication Training (Douglas et al., 2018)	Communication	6 modules; completion 1-7 days.	Slides with visuals and narration, video demonstrations/models of strategies, demonstration scenarios, and quizzes. Access to paper module handouts and instructions.	Self-directed. Post-training: able to ask questions; prompted to use strategies; no other coaching provided.
ImPACT Online (Ingersoll et al., 2016; additional information in: Ingersoll & Berger, 2015; Pickard et al., 2016)	Social communication skills	12 lessons (approx. 80 min each); 1 lesson / week; 6 months' access.	Narrated slideshow with embedded video clips of expert therapist or parent demonstrating step-by-step techniques; written manual; self-check quizzes; interactive exercises with immediate feedback (e.g., watch video clips and identify correct use of intervention techniques); homework plan; reflective questions. Supplemental material: video library, resource page links, moderated parent forum, weekly email tips.	Self-directed group: technical support only. Therapist-assisted group: 2 x 30-min remote coaching sessions / week with trained therapist (clarified content, provided live feedback).
Pivotal Response Treatment (PRT) training program (McGarry et al., 2019)	Social communication skills	6 learning modules (15-30 minutes); 6-week duration.	Information slides; video examples modelling proper strategy use; quizzes to assess comprehension with automatic feedback; module summary sheet; parents required to submit 5 min. video of PRT	Self-directed.

Intervention, by NDD	Target of Intervention	# Sessions / Duration	Online Content Delivery	Level of Support
			implementation with child after each module.	
Reciprocal Imitation Training (RIT) (Wainer & Ingersoll, 2013 – sample 2; Wainer & Ingersoll, 2015)	Imitation	*Pilot*: 5 modules. *Newer*: 4 lessons.	*Pilot version* (2013)*:* PPTs with audio lectures, video examples, short comprehension quizzes with immediate feedback, interactive learning tasks (judge video clips for accurate use of techniques). Downloadable PDF manual available. *Newer version* (2015): Animated slideshow with audio lectures; interactive learning tasks (judge video clips); homework plan; reflection questions. Access to video library and resources page.	Self-directed during online training phase(s). In Wainer & Ingersoll (2015), coaching condition: 3 x 30 min remote coaching sessions (answer questions / provide feedback).
Enhancing Interactions (Ibanez et al., 2018)	Engagement in routines	3-module tutorial (~ 6 hrs to complete entire tutorial). Encouraged to complete over 4-5 sessions; 1 month access.	Interactive learning activities, video demonstrations, ability to customize tutorial content & choose activities within each routine to work on.	Self-directed.
ABA Web-Based Training Program (Blackman et al., 2019)	Parent-child interactions; parent knowledge	6 modules (30-40 min long); 1 / week; 6-week duration.	Pre-recorded researcher-narrated PPT presentation; video models of strategies; active parent responding questions (i.e., multiple choice online quiz) with correct answers shown. Self-paced (pause / resume	Self-directed.

Intervention, by NDD	Target of Intervention	# Sessions / Duration	Online Content Delivery	Level of Support
			at later time). Must complete each module to continue to next.	
Project CHASE (Children with Autism Supported to Exercise) (Healy & Marchand, 2019)	Physical activity	N/A – Facebook posts rather than sessions. 4-week duration.	Hosted within private Facebook Group; facilitators posted 6 intervention components in group: physical activity promotion materials, discussion prompts for participants, interactive sharing of social support, motivational messages, rewards. Google Forms used for parents to monitor child's physical activity.	Self-directed engagement with Facebook. Motivational reminders sent 1x week via Facebook Messenger.
Parent Sleep Education (Roberts, Smith, & Sherman, 2019)	Child sleep behaviours; parent quality of life and fatigue	2 x 2-hr online podcast sessions (separated by 1-2 weeks) + blogging.	Online Moodle podcasts that incorporated PPT presentation with voiceover slides, videos demonstrating strategies. Discussion questions and blog entries.	Self-directed. Able to email questions to instructor. Instructor posted discussion questions for blogs.
ADHD				
Blended Behavioural Parent Training (BPT) (Breider et al., 2019)	Externalizing behaviour problems	6 modules; 20-week duration.	Theory, exercises, online assignments. Must demonstrate understanding to continue to next module.	Self-directed modules + online therapist feedback + supportive face-to-face parent-therapist contacts (90 min introductory contact, 4 x 45-60 min evaluation contacts; follow-up contact). Therapists scheduled

Intervention, by NDD	Target of Intervention	# Sessions / Duration	Online Content Delivery	Level of Support
				extra contact if they felt parents needed it.
Promoting Engagement for ADHD pre-Kindergartners (PEAK) (Dupaul et al., 2018a, 2018b)	ADHD symptoms and behaviours	10 online sessions (1 session / week); 10-12 weeks access.	PPT slides with narration; video examples of strategies; check-in questions for parents about strategy usage; homework assignments / practice.	Weekly phone calls with consultant / research assistant to review strategies / answer questions.
Triple P Online (TPOL) (Franke et al., 2017; Day & Sanders, 2018)	Child behaviour problems and ADHD symptoms; negative parenting styles	8 modules; 1 module / week; 4 months access.	Multimedia videos to demonstrate strategies; interactive activities; dynamically generated workbook for tracking program progress; ability to set goals and develop prevention plan. Optional communication tools: text reminders.	*Franke et al. (2017)*: Self-directed + 2x phone consultations with facilitator to tailor strategies / problem-solve. *Day & Sanders (2018):* Self-directed condition. Technical support available. Phone-supported condition: 1x consult/ week.
FASD				
FASD Education and Training for Behavioural Regulation	Child behavioural regulation;	Single web page.	Web page containing graphics, PPT slide show that incorporated written text information.	Self-directed. Technical support available.

Intervention, by NDD	Target of Intervention	# Sessions / Duration	Online Content Delivery	Level of Support
(Kable et al., 2012)	parent knowledge			

Table 2.4

Summary of research studies evaluating online parent-implemented interventions and quality ratings, by primary NDD group.

Intervention	Design	WLC (Y/N)	Participants (*n*, child age mean or range)	Comorbidities (included / excluded)	Parent Characteristics	Child Outcomes	Parent Outcomes	Results	Effec-tive (Y/N)	Quality Rating (/27, %age)
ASD										
1. POWR Online Communication Training (Douglas et al., 2018)	SC, MP	N	*n* = 3; mean age = 4.3y	Develop-mental delay.	Mothers; mean age = 37 y; English speaking; all had university experience.	-Communication$_1$	-Parent-provided communication opportunities$_1$ -Responses to child communication$_1$	Medium effects: increased child communication & parent-provided communication opportunities.	Y	15, 55.6%
2. ImPACT Online (Ingersoll et al., 2016)	RCT (pilot)	N	*n* = 27 (self-directed 13, therapist-assisted 14). Age range: 27-73. Mean age (SD): self-directed = 46.1m (13.2); therapist-assisted = 41.6m (12.2).	No information.	92–100% female; 36 – 54% had less than college degree; 29 – 46% not employed; 64-77% resided in underserved area.	-Language (spoken during parent-child interaction)$_1$ -Expressive vocabulary (MCDI) -Adaptive functioning (VABS-II)	-Parenting competence (PSOC) -Family impact (FIQ) -Intervention fidelity$_1$	Both groups improved child language, parent intervention fidelity, self-efficacy, stress, and positive perceptions. Child social skills only improved in therapist-assisted; greater gains in therapist-assisted.	Y	20, 74.1%
3. Pivotal Response Treatment (PRT) training program	PP	N	*N* = 11; mean age (SD) = 36.8m (8.1), range = 20-46m.	Excluded comorbid medical / psychiatric disorders. No	72.7% female, 27.27% male; 90% married; all had bachelor's	-Social communication behaviours$_1$	-Parent PRT fidelity$_1$ -Social validity (satisfaction, confidence,	Improved child communication behaviours and parent treatment fidelity.	Y	17, 63%

Intervention	Design	WLC (Y/N)	Participants (*n*, child age mean or range)	Comorbidities (included / excluded)	Parent Characteristics	Child Outcomes	Parent Outcomes	Results	Effec-tive (Y/N)	Quality Rating (/27, %age)
(McGarry et al., 2019)				further information on comorbidities	degree or more education.		perceptions of child behaviour change)$_2$			
4. Reciprocal Imitation Training (RIT) a. Wainer & Ingersoll (2013; sample 2 only)	SC, MB	N	*n* = 3 dyads; child mean age = 61m, range = 26-88m.	No information.	Mothers; no previous training in naturalistic behavioural intervention techniques; 2 had graduate degrees / 1 had some college.	-Imitation$_1$	-Program utilization -Knowledge$_2$ -Treatment fidelity$_1$ -Treatment acceptability (modified BIRS)	Substantial increases in imitation rates for 2 children; parent knowledge increase for all.	Y	15, 55.6%
b. Wainer & Ingersoll (2015)	SC, MB	N	*n* = 5 dyads; child age range = 29-59m.	No information.	Remote location; diverse cultural backgrounds; 4 graduate degrees, 1 had some college; 80% married; no previous formal parent training; required access to internet.	-Imitation$_1$	-Program engagement -Knowledge$_2$ -Treatment fidelity$_1$ -Treatment acceptability (modified BIRS)	Self-directed phase: strong improvement in parent knowledge, increased treatment fidelity, small increase in imitation. Coaching phase: continued increases in knowledge, fidelity, imitation. Overall clinical significance unknown.	N	16, 59.3%
5. Enhancing Interactions	RCT	Y	*N* = 104 (tutorial 52, control 52)	Excluded severe auditory,	90.4% mothers; 90.4% white; mean age:	-Engagement behaviours during	-Strategy usage (parent behaviour survey)	Increased parent strategy use, child engagement,	Y	19, 70.4%

Intervention	Design	WLC (Y/N)	Participants (*n*, child age mean or range)	Comorbidities (included / excluded)	Parent Characteristics	Child Outcomes	Parent Outcomes	Results	Effec-tive (Y/N)	Quality Rating (/27, %age)
(Ibanez et al., 2018)			Age range: 18-60m; mean age (SD): tutorial = 42.8m (13.4), control = 44.8m (12.5)	visual or motor impairments. No further information on comorbidities.	34.71 y mothers, 34.61 y fathers. 38.4% no college degree, 46.1% had college degree, 15.4% graduate degree. Required access to internet.	routines (parent-reported)$_{2,3}$ -Social communication skills (PIA-CV)$_{3}$	-Stress (PSI-SF) -Parenting efficacy (PES) -Usability & satisfaction (SUS + USQ$_{2}$)	parenting efficacy, and lower parent stress in tutorial group compared to control.		
6. ABA Web-Based Training Program (Blackman et al., 2019)	PP	Y	*n* =18 dyads (online *n* = 6; in-vivo *n* = 7; control *n* = 5) Mean age: 4.4y (online 4.8y, in vivo 3.5y, control 4.8y)	No information.	77.7% female, 61.1% white, 33.3% had some college, 33.3% had college degree, 16.7% had advanced degree. 33.3% single, 55.6% married, 11.1% divorced. Excluded prior training in ASD / ABA.	-Parent-child interactions$_{1}$	-Knowledge$_{2}$ -Stress (PSI-SF) -Parental competence (PSCS)	Parent-child interaction and parent knowledge improved for both groups; no significant differences between groups.	Y	15, 55.6%
7. Project CHASE (Healy & Marchand, 2019)	PP	N	*n* = 13 Mean age (SD): 9.4y (3.1); range = 6-16y.	53.8% had ADHD; 23.1% had speech delay / apraxia. Other comorbidities:	92.3% female; 84.6% Caucasian; mean age 41.3 y; 70% used Facebook	- Physical activity levels (GLTQ)$_{3}$	-Feasibility -Study engagement	Increased parent-reported physical activity for all participants; not statistically significant.	N	15, 55.6%

Intervention	Design	WLC (Y/N)	Participants (*n*, child age mean or range)	Comorbidities (included / excluded)	Parent Characteristics	Child Outcomes	Parent Outcomes	Results	Effec-tive (Y/N)	Quality Rating (/27, %age)
				SPS, dyspraxia, LD, ID, epilepsy, anxiety.	several times a day. Education: high school 30.8%, associate degree 46.2%, bachelor degree 15.4%, master's degree 7.7%. Recruited from a remote county with a high poverty level.					
8. Parent Sleep Education (Roberts et al., 2019)	PP	N	*n* = 25 (F2F 13, online 10). Age range: 4-12y. Mean age (SD): F2F = 8.6y (1.8), online = 7 (2.6).	47.8% had ADHD; 30.4% had anxiety.	91.3% female; English-speaking; mean age: mothers 36:8 y, fathers 38:5 y; 73.9% married / partnered; approx. half attended college or earned degree.	-Sleep behaviours (CSHQ; actigraphy)	-Quality of life (PedsQL) -Fatigue (FAS) -Satisfaction[2]	Total sleep score & parent quality of life improved for both F2F and online; online group had improved / sustained parent fatigue and decreased night wakings.	Y	18, 66.7%
ADHD										
9. Blended BPT (Breider et al., 2019)	RCT (pilot)	N	*n* = 22 parents (online 11, F2F 11). Mean age (SD): online = 7.5y	Disruptive behaviour disorder, Tourette's syndrome,	Required both parents to participate and availability of computer.	-Behaviour problems severity (Dutch version of CBCL –	-Attrition -Satisfaction[2]	Blended online parent training inferior to F2F training.	N	18, 66.7%

Intervention	Design	WLC (Y/N)	Participants (*n*, child age mean or range)	Comorbidities (included / excluded)	Parent Characteristics	Child Outcomes	Parent Outcomes	Results	Effec-tive (Y/N)	Quality Rating (/27, %age)
			(2.2), F2F = 8.1y (1.9); range = 4-12 y.	anxiety. No restrictions on comorbidities. IQ must be > 70.	Excluded prior behavioural parent training. Educational levels: most parents in ‘middle’ (54-60%) or ‘high’ (27-10%).	Externalizing Scale)[3]				
10. Triple P Online (TPOL) a. Day & Sanders, 2018	RCT	Y	*n* = 183 (TPOL 57, telephone supported TPOLe 66, WLC 60). Mean age (SD): 3.5y (1.5); TPOL = 3.4y (1.5), TPOLe = 3.6y (1.7), WLC = 3.4y (1.2).	Excluded ID, Developmental Delay, ASD. No further information on comorbidities.	96.1% mothers; mean age: 34.9 y (range 22 – 51y); 93.4% white; 60.6% had university degree; 14% had financial difficulty; 78.6% reported clinical levels of conflict with partner around parenting. Met at least one socioeconomic risk factor associated with higher levels of child socio-emotional & behavioural problems. Must	-Behaviour problems (ECBI) -Behaviour patterns over 24 hour period (modified PDR)[3]	-Parenting practices / styles (PS) -Parent adjustment & mental health (DASS) -Parenting confidence (PTC) -Relationship quality & adjustment (PPC, RQI) -Parental anger (PAI) -Program engagement -Parent Satisfaction (CSQ)	Telephone-supported (TPOLe) outcomes better than self-directed (TPOL) – greater improvements in negative parenting style & intensity of difficult child behaviours.	Y	21, 77.8%

Intervention	Design	WLC (Y/N)	Participants (*n*, child age mean or range)	Comorbidities (included / excluded)	Parent Characteristics	Child Outcomes	Parent Outcomes	Results	Effec-tive (Y/N)	Quality Rating (/27, %age)
					not be receiving other parent support.					
b. Franke et al., 2017	RCT (pilot)	Y	*n* = 53 (intervention 27, delayed intervention 26). Mean age = 4y; range = 3-4y.	Excluded developmental disorders. No further information on comorbidities.	Mothers mean age 35.4 y; fathers (*n* = 43) mean age 38.8 y; 55.7% parents had university degree. 32% of families had one parent with clinically elevated levels of ADHD symptoms.	*Parent-report:* -Behaviour (Conners EC-BEH-P) *Teacher report:* -Behaviour & peer relations (CBS) -Hyperactivity & peer problems (SDQ)	-Parenting practices / styles (PS, PSDQ) -Mental health (DASS) -Parenting competence (PSOC) -Satisfaction (CSQ)	Improvements in parent-rated ADHD symptoms, social functioning, teacher-rated prosocial behaviour, and parenting outcomes.	Y	21, 77.8%
11. Promoting Engagement for ADHD pre-Kindergartners (PEAK) a. DuPaul et al., 2018b – Step 5 sample	PP	N	*n* = 8 Age range = 3:0-5:11y.	Excluded other primary disabilities (e.g., ASD) or global ability score < 80 on DAS-II. No further information on comorbidities.	No information provided.	-ADHD symptoms & behaviours (CERS)[3]	-Acceptability & feasibility (IRP-15, focus group discussion)	ADHD symptoms / behaviours reduced; post-treatment ratings still in clinical range for most.	Y	12, 44.4%
b. DuPaul et al., 2018a	RCT (pilot)	Y	*n* = 47 (online = 15; F2F = 16; WLC = 16)	53.2% had comorbid ODD; 6.4% had comorbid ODD + CD.	Parent education: 40.4% completed postgrad,	-ADHD symptoms & behaviours (CERS)[3]	-Attendance/ completion -Knowledge[2] -Stress (PSI-SF) -Treatment fidelity	Reduced ADHD symptoms, improved mood/affect, & greater parent	Y	17, 63%

Intervention	Design	WLC (Y/N)	Participants (*n*, child age mean or range)	Comorbidities (included / excluded)	Parent Characteristics	Child Outcomes	Parent Outcomes	Results	Effec-tive (Y/N)	Quality Rating (/27, %age)
			Mean age (SD) = 4.5y (.6); range = 3:0-5:11y.	Excluded ASD, PDD, ID, neurological damage, significant motor or physical impairments, global ability score < 80 on DAS-I.	21.2% had college degree, 27.6% attended college. Required electronic device with internet access.		-Treatment acceptability (modified IRP-15)	knowledge in treatment groups compared to control. No significant differences between F2F and online.		
FASD										
12. FASD Education and Training for Behavioural Regulation (Kable et al., 2012)	RCT	Y	*n* = 59 (internet 18, F2F 23, control 18). Mean age (SD): internet 7.3y (2.7), F2F 6.7y (3.4), control 6.2y (2.9).	Experienced CPS involvement: 66.7 – 83.3%; physical abuse: 25 – 28.6%; sexual abuse: 6.3 – 18.2%. No further information on comorbidities	Length of caregiver education:13.8 – 14.5 y.	-Behaviours (CBCL)[3]	-Satisfaction[2] -Knowledge[2]	Both treatment groups improved in knowledge, behavioural skills, and advocacy compared to control; only F2F group produced changes in child behaviour. Internet group not more effective than F2F.	Y	18, 66.7%

Notes:
[1] = coded / scored by researchers. [2] = author-made measure / questionnaire. [3] = parent-reported. ASD = Autism Spectrum Disorder; ADHD = Attention-Deficit / Hyperactivity Disorder; CD = Conduct Disorder; FASD = Fetal Alcohol Spectrum Disorder; ID = Intellectual Disability; LD = Learning Disability; NDD = Neurodevelopmental Disorders; ODD: Oppositional Defiant Disorder; PDD = Pervasive Developmental Disorder; PDD-NOS = Pervasive Developmental

Disorder – Not Otherwise Specified; SPD = Sensory Processing Disorder. DSM-IV-TR: Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision. DSM-5: Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition. RCT = Randomized Controlled Trial. SC = Single-case design. MP = Multiple probe design. MB = Multiple baseline design. PP = Pre-test / post-test design. WLC = Wait list control. F2F = Face-to-face. PRT = Pivotal Response Treatment. RIT = Reciprocal Imitation Training. SSIG = Statistically Significant. Y = Yes. N = No. Outcome measure abbreviations: ADOS-2 = Autism Diagnostic Observation Schedule, Second Edition. ADOS-G = Autism Diagnostic Observation Schedule – Generic. BIRS = Behavioral Intervention Rating Scale. CBCL = Child Behavior Checklist. CBS = Child Behavior Scale. CERS = Conners Early Childhood Rating Scale. CES-D = Centre for Epidemiological Studies – Depression Scale. Conners EC-BEH-P = Conners Early Childhood Behavior – Parent. CSHQ = Children's Sleep Habits Questionnaire. CSQ = Client Satisfaction Questionnaire. DASS = Depression Anxiety Stress Scales. ECBI = Eyberg Child Behavior Inventory. FAS = Fatigue Assessment Scale. FIQ = Family Impact Questionnaire. GLTQ = Godin Leisure-Time Questionnaire. IRP-15 = Intervention Rating Profile-15. MCDI = McArthur-Bates Communicative Development Inventory. PAI = Parental Anger Inventory. PDR = Parent Daily Report. PedsQL = PedsQL Inventory (Family Impact Module 2.0) Parent Report. PES = Parental Efficacy Scale. PIA-CV = Parent Interview for Autism – Clinical Version. PPC = Parent Problem Checklist. PS = Parenting Scale (Arnold, O'Leary, Wolff, & Acker, 1993). PSCS = Parenting Sense of Competence Scale (Gibaud-Wallston & Wandersmann, 1978). PSDQ = Parenting Styles and Dimensions Questionnaire. PSI-SF = Parenting Stress Index – Short Form. PSOC = Parenting Sense of Competence (Johnson & Mash, 1989). PTC = Parenting Tasks Checklist. RQI = Relationship Quality Index. SDQ = Strengths & Difficulties Questionnaire. SUS = System Usability Scale. USQ = User Satisfaction Questionnaire. VABS-II = Vineland Adaptive Behavior Scales, Second Edition. DAS-II = Differential Ability Scales-II.

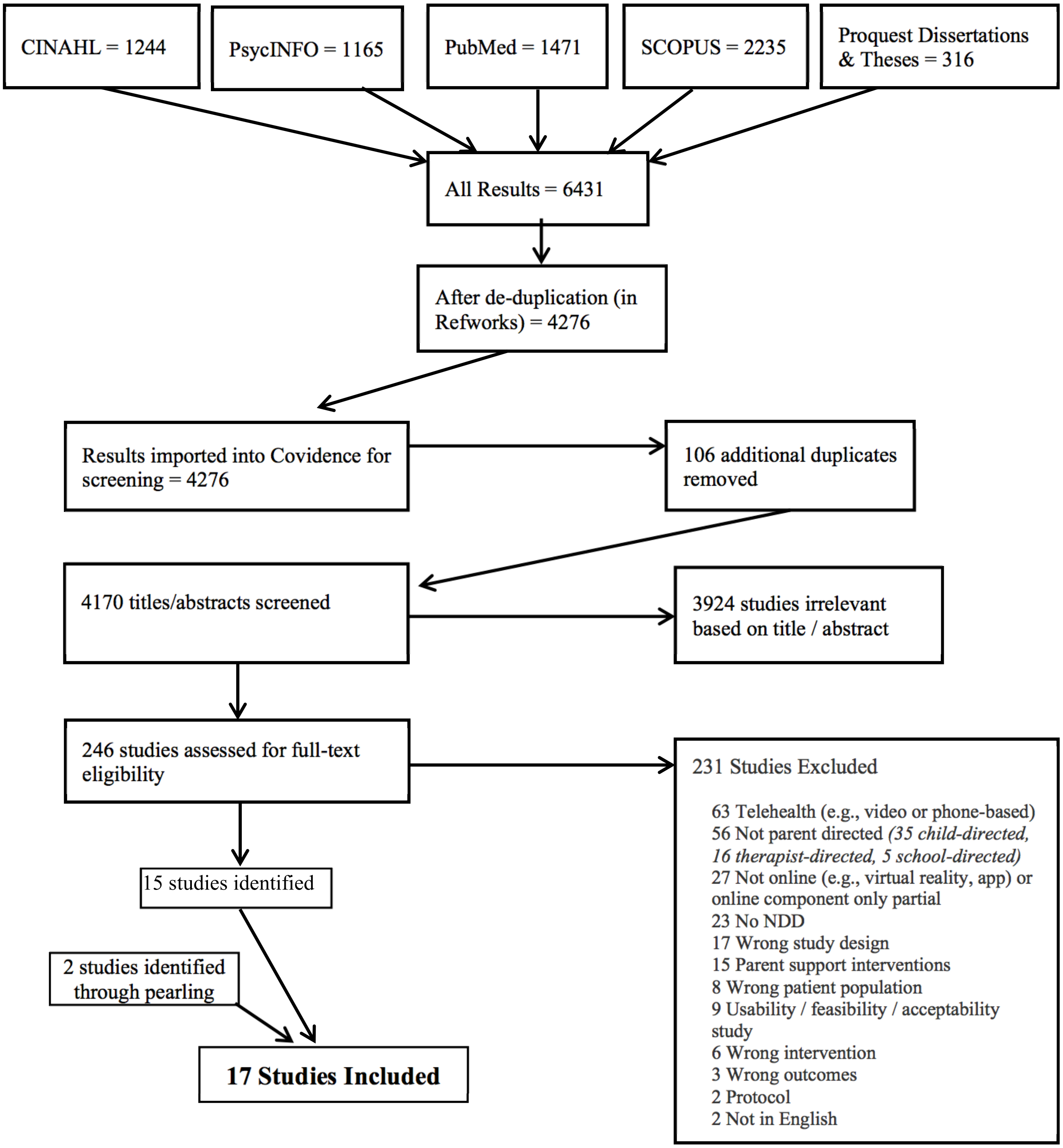

Figure 2.1. *PRISMA Diagram*

Chapter 3: Barriers and Facilitators to Treating Insomnia in Children with Autism Spectrum Disorder and Other Neurodevelopmental Disorders: Parent and Health Care Professional Perspectives

The manuscript based on this experimental study is presented below. Readers are advised that Kim M. Tan-MacNeill, under the supervision of Dr. Penny Corkum and Dr. Isabel Smith, was responsible for developing the study protocol and applying for ethical approval; reviewing extant literature and formulating research questions; training and supervising research staff; completing and overseeing data collection; completing all analyses; and all aspects of the writing process. Colleagues Anastasija Jemcov and Laura Keeler were involved with aspects of the data collection and qualitative analysis process, under Kim's supervision. This research was completed in consultation with dissertation committee members Dr. Jill Chorney and Dr. Shannon Johnson, who provided editorial feedback. Editorial feedback was also provided by members of the *Better Nights, Better Days for Children with Neurodevelopmental Disorders* (BNBD-NDD) research team: Dr. Shelly K. Weiss, Dr. Cary A. Brown, Dr. Evelyn Constantin, Dr. Roger Godbout, Dr. Ana Hanlon-Dearman, Dr. Osman Ipsiroglu, Dr. Graham J. Reid, Dr. Sarah Shea, and Dr. Esmot Ara Begum.

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Abstract

Background/Aims: Insomnia is highly prevalent in children with neurodevelopmental disorders (NDDs), yet little research exists on sleep treatment access, utilization, and provision in this population. This study explores barriers and facilitators to access, use, and provision of treatment for sleep problems as experienced by parents of children with NDDs, including Autism Spectrum Disorder (ASD), Attention-Deficit/Hyperactivity Disorder (ADHD), Cerebral Palsy (CP) and Fetal Alcohol Spectrum Disorder (FASD), and health care professionals who work with children with these conditions.

Method: Transcripts from online focus groups and interviews, conducted separately with parents of children with NDDs (*n*=43) and health care professionals (*n*=44), were qualitatively analyzed using content analysis for key themes.

Results: Barriers included limited access to/availability of treatment, lack of knowledge/training, NDD-specific factors (e.g., symptoms, medications, and comorbidities), parent factors (e.g., capacity to implement treatment, exhaustion), and the challenging, intensive nature of sleep treatment. Facilitators included positive beliefs and attitudes, education, support, and ability to modify treatments for NDD symptoms. Barriers and facilitators were similar across all four NDDs.

Conclusions: Results highlight a need for more education about sleep in NDDs and to develop accessible interventions, as well as the potential of a transdiagnostic approach to sleep treatment in this population.

Introduction

Background

Neurodevelopmental disorders (NDDs) emerge in early childhood and are linked to disturbances in central nervous system functioning, which can cause impaired cognition, communication, motor skills, and/or behaviour, and functional impairment in a variety of daily life domains (American Psychiatric Association, 2013). Sleep problems are highly prevalent in children with NDDs, with rates ranging from 40 – 86% (Robinson-Shelton & Malow, 2016; Romeo et al., 2014). Insomnia, the most common sleep problem experienced by children with NDDs, includes difficulty falling and staying asleep (American Academy of Sleep Medicine, 2014). Throughout this paper, we will use the terms insomnia and sleep problems interchangeably.

Sleep problems have been shown to increase the severity of NDD symptoms as well as behavioural and emotional problems, and to have negative effects on children's daytime functioning (Tudor, Hoffman, & Sweeney, 2012; Goldman, McGrew, Johnson, Richdale, Clemons, & Malow, 2011; Newman, O'Regan, & Hensey, 2006). Children's sleep problems occur within a broad psychosocial context and may affect the whole family; for example, parents of children with NDDs and sleep problems experience high levels of stress (Doo & Wing, 2005).

Development of effective treatments for insomnia in children with NDDs is important, given the high prevalence and negative effects of sleep problems. Behavioural interventions are the first-line recommendation for pediatric insomnia in both NDD and typically developing (TD) populations (Malow et al., 2012). Research on effective sleep interventions for children with NDDs is expanding, with several recent randomized controlled trials (RCTs) (e.g., Hiscock, Sciberras, & Mensah, 2015). A recent systematic review found support for a transdiagnostic behavioural approach to treating sleep problems in children with NDDs (Rigney et al., 2018), wherein the same behavioural treatment principles are applied across multiple diagnoses, with minor modification of strategies originally developed for TD children (e.g., psychoeducation, healthy sleep practices, extinction).

Emerging research suggests that access to and uptake of behavioural sleep interventions by families of children with NDDs is limited (e.g., Bessey, Coulombe, Smith, & Corkum, 2013; Boerner, Coulombe, & Corkum, 2014). Additionally, front-line health care professionals (HCPs) are generally not well trained to provide sleep interventions (e.g., Boerner et al., 2014), much less for special populations such as children with NDDs. As such, we need to explore factors influencing families' seeking of, access to, and uptake of treatment for sleep problems in their children with NDDs, as well as the factors influencing HCPs' ability to provide such treatment. This information will provide a foundation for the development of effective sleep interventions for this population.

We employed focus groups (or interviews when participants were not able to attend focus groups) to gather the perspectives of parents of children with NDDs and HCPs on barriers and facilitators to access, uptake, and provision of sleep treatments for children. Four prevalent NDDs that encompass a range of symptoms and functional impairments were included: Attention-Deficit/Hyperactivity Disorder (ADHD), Autism Spectrum Disorder (ASD), Cerebral Palsy (CP), and Fetal Alcohol Spectrum Disorder (FASD). The results of the study will identify unmet needs in the areas of treatment delivery and use, accessibility, and professional development and training in order to inform the development of a sleep intervention for children with these four NDDs.

Research Objectives

The research objectives were to explore the barriers and facilitators experienced by 1) parents, in seeking, accessing, utilizing, and implementing treatments for sleep problems in children with NDDs; and 2) HCPs, in their access to information about and provision of sleep treatments for children with NDDs. We predicted that lack of knowledge, training, and time may be barriers reported by HCPs. We expected that both parents' and HCPs' beliefs and attitudes about the nature of sleep problems in NDDs and their treatability would influence responses regarding treatment seeking, access, uptake, and provision.

Method

Participants

This study was approved by the Research Ethics Board of the IWK Health Centre in Halifax, Nova Scotia, Canada. Informed consent was obtained from all participants, who were recruited online via social media, through sharing of recruitment advertisements by NDD-related parent and health organizations, and through the authors' professional networks. Conducting individual interviews became necessary for some participants, due to difficulty accommodating time zones and schedules.

Parent Participants. The final sample included 43 parents or caregivers (hereafter, parents) of children aged 4 – 12 years with parent-reported diagnoses of ADHD (*n* = 9), ASD (*n* = 20), CP (*n* = 6), and/or FASD (*n* = 8) from a physician or psychologist, as well as behavioural sleep problems confirmed by a screening questionnaire. In cases where children had comorbid ADHD with ASD, CP, or FASD, the ASD/CP/FASD diagnoses were considered primary for assigning them to a disorder group (e.g., comorbid FASD and ADHD = FASD group). As such, children of parents in the ADHD group could not have comorbid ASD, CP, or FASD. Parents were required to live in Canada, have access to a computer, internet, web-camera and microphone (or telephone), and be comfortable speaking/reading English. Parent-reported formal diagnoses of sleep disorders other than insomnia (e.g., sleep apnea) were an exclusion criterion. Information about children's comorbid diagnoses (e.g., NDD, neurological, physiological, mental health) and medication use was recorded but not used as exclusionary criteria.

Figure 3.1 depicts parent participation, and Table 3.1 contains demographic information. Twenty-seven parents participated in focus groups and 16 parents completed individual interviews. Most parents were biological mothers (74.4%). The mean age of parents was 38.5 years (SD = 7.1, range = 25-65), and most parents were married/common-law (*n* = 33, 75.7%). Most lived in cities (*n* = 26, 60.4%), were of Caucasian heritage (*n* = 39, 90.7%), and had completed high school and some post-secondary education (*n* = 39, 90.7%). The average reported number of other children in the home was 1.95 (SD = .9, range 1 – 5). Most parents were from Ontario (n = 17, 39.5%), British Columbia (n = 8, 18.6%), and Alberta (n = 7, 16.3%), with the remainder from Nova Scotia (*n* = 4), New Brunswick (*n* = 2), Newfoundland and Labrador (*n* = 2), and Prince Edward Island, Manitoba, and Quebec (each *n* = 1).

Most children were male (n = 29, 67.4%) and mean age was 8.5 years (SD = 2.5, range 4.3 – 12.6). Most children had at least one other parent-reported diagnosis (*n* = 30, 69.8%), including other comorbid NDDs (ASD, ADHD, CP, or FASD; *n* = 9, 20.1%) or mental health diagnoses (*n* = 26, 60.5%); anxiety was common (*n* = 16, 37.2%). Children also had a range of parent-reported physical health conditions (*n* = 19, 44.2%), most frequently epilepsy/seizure disorders (*n* = 5, 11.6%), other neurological disorders (*n* = 7, 16.3%), gastrointestinal disorders (*n* = 6, 14%), and respiratory disorders (*n* = 4, 9.3%).

In terms of behavioural insomnia (Anders & Dahl, 2007), fifteen (34.9%) children met criteria for bedtime resistance/sleep onset problems, six (14%) met criteria for night waking problems, and 18 (41.9%) met criteria for both. Four children (9.3%) were below threshold for behavioural insomnia, but were included as their parents reported high severity/impact of sleep problems. Twenty parents (46.5%) reported that their children woke too early in the morning. Frequently reported problems were: problems falling asleep (*n* = 38, 88.4%), lying awake in bed after lights out for more than 20 minutes (*n* = 38, 88.4%), problems staying asleep (*n* = 34, 79.1%), getting out of bed once expected to stay in bed for the night (*n* = 32, 74.4%), and waking during the night with difficulty falling back asleep (*n* = 32, 74.4%).

Health Care Professional Participants. The final sample included 44 credentialed Canadian HCPs who practiced with 4- to 12-year-olds with NDDs. As many HCPs practiced with more than one NDD group, they were asked to choose the NDD with which they worked most often for the focus group/interview. The breakdown of HCPs by NDD was as follows: ADHD (*n* = 8), ASD (*n* = 21), CP (*n* = 8), and FASD (*n* = 7). Eligible professions for participation included physicians, psychologists, nurses, social workers, occupational therapists, and Board-Certified Behaviour Analysts (BCBAs; certified behaviour analysts who primarily work with children with ASD and provide behavioural interventions). Figure 3.2 depicts HCP participation. HCPs required access to a computer/internet, web camera and microphone (or telephone), and fluency in English. To ensure a diverse sample of HCPs, no minimum percentage of practice time was specified for working with children with NDDs or with sleep problems.

Twenty-one HCPs participated in focus groups, whereas 23 participated in individual interviews. Table 3.2 shows demographic information. Professions included occupational therapists (*n* = 15), clinical psychologists (*n* = 10), general paediatricians (*n* = 1)/developmental paediatricians (*n* = 6), nurses (*n* = 4), BCBAs (*n* = 4; ASD only), family physicians/general practitioners (*n* = 2), and social workers (*n* = 2). The majority of HCPs were from Ontario (*n* = 16, 36.4%) and Nova Scotia (*n* = 13, 29.5%), followed by Alberta (*n* = 5, 11.4%), British Columbia (*n* = 4, 11.4%), New Brunswick (*n* = 3, 6.8%), and Quebec (*n* = 3, 6.8%). Most HCPs were female (*n* = 41, 93%), had a Master's or higher degree (*n* = 31, 70.5%), and practiced primarily in healthcare settings (*n* = 27, 61.4%), most commonly in hospitals (*n* = 14, 31.8%). HCPs averaged 13.9 years of experience working with children (SD = 9.9, range 2 – 35 years); most specialized in working with children with NDDs (*n* = 34, 77.3%).

Screening, Eligibility, Demographic, and Background Information Measures

Parents. Two-step screening was completed online: 1) Parents completed an author-made questionnaire targeting inclusion and exclusion criteria (Appendix B). 2) Parents who met initial inclusion/exclusion criteria then completed a questionnaire consisting of general diagnostic information, the Behavioural Insomnia Questionnaire

(BIQ; Anders & Dahl, 2007; modified by authors) to assess the presence of behavioural sleep problems, and the first six items of the Pediatric Sleep Questionnaire (PSQ; Chervin, Hedger, Dillon, & Pituch, 2000) to screen for sleep apnea (Appendix C). The BIQ provides a cut-off score to determine presence of sleep onset and night-waking problems over the previous month; author additions included parent ratings of the perceived severity and impact of their children's sleep problems across multiple domains (e.g., school, fatigue, family life), as well as reports of co-sleeping. Eligible parents then completed a Demographic Information Questionnaire (author-developed; based on Canadian census).

***Health Care Professionals*.** Health care professionals completed an author-made questionnaire that asked about inclusion and exclusion criteria, identifying their professional group and the NDD group(s) with which they worked (Appendix D). Eligible HCPs completed a Health Care Professionals' Demographic Information and Training Questionnaire (author-adapted from measures in Meltzer, Phillips, & Mindell, 2009), which collected information on HCPs' professional practice with NDDs, sleep-related training/education, practice setting, and self-rated competence in treating sleep problems.

Focus Groups and Interviews

After eligible parents and HCPs were enrolled in the study and scheduled for a focus group session or interview, they were instructed in using the video-conferencing software and required to test the software prior to participation.

Description of focus groups/interviews. Separate focus groups and interviews were held for parents (10 focus groups, 16 interviews) and HCPs (8 focus groups, 22 interviews). Groups/interviews were separated by NDD (e.g., ASD-only parent focus group). Within HCP focus groups, HCPs of different disciplines were combined. Groups/interviews were conducted using encrypted video-conferencing software (Blackboard Collaborate/Collaborate Ultra) that displayed PowerPoint slides showing discussion questions for the participants. A minority of participants (parent $n = 5$, HCP $n = 7$) required teleconferencing (i.e., integrating a phone without video into the software) due to technical difficulties. One local HCP was interviewed and recorded in-person (at their request). Due to software constraints, the present study set a maximum of 5 participants per group plus a moderator, which is consistent with online focus group guidelines (Tuttas, 2014). Each focus group (approximate duration 1.5 hours) was facilitated by the first author (K.T.M.). Volunteer research assistants acted as second moderators and were available for technical support during focus groups. All interviews (approximate duration 1 hour) were conducted solely by the first author using the same software as the focus groups.

Topic guides. Semi-structured topic guides (Appendices E and F) for focus groups/interviews focused on the experience of treatment, from seeking to implementing. Parent topics included knowledge of sleep in children with NDDs, experience of seeking treatment for insomnia, uptake/use of treatments (separated into medications, over-the-counter treatments such as melatonin/natural remedies, and behavioural treatments). HCP topics included familiarity with and extent of involvement with sleep treatment for children with NDDs, knowledge about and access to sleep treatments, and provision of sleep treatment. At the end of each session, participants were asked what they felt was the most important issue discussed and if anything had been missed. Participants were not asked to review transcripts.

Analysis

Focus group/interview sessions were audio-/video-recorded, transcribed, and de-identified. Transcripts were analyzed in NVivo software (QSR International, NVivo for Mac, version 12.4.0), using qualitative content analysis (Schreier, 2012). The first author (K.T.M.) developed separate coding frames for parents and HCPs in consultation with authors I.S. and P.C. and trained a second coder (L.K.). Transcripts were reviewed and recoded multiple times to ensure coding agreement and that the coding frames were suitable. Parent and HCP data were coded separately.

As transcripts were reviewed, the smallest units of analysis that contained a coherent meaning (typically a sentence, group of sentences, or a single response from a participant) were identified as separate codes. Given the complexity of responses, some sections of text yielded several different codes. Following the initial round of coding to identify individual barrier and facilitator codes, the codes were grouped into broader themes and sub-themes. These themes constituted the final barriers and facilitators and are presented in Tables 3.3 – 3.6. Frequency data are available upon request. To examine group differences, complete lists of codes and frequencies were generated for all parent data and all HCP data respectively, then separate lists were generated for each NDD group (e.g., parents – ASD, ADHD, CP, FASD). Similarities and differences were noted in the presence of codes across NDD groups (within parent or HCP data overall).

Results

Parents

Barriers. Four barriers were identified for parents, consisting of 34 individual codes (see Table 3.3): 1. Access to and Availability of Services, 2. Experience with Service (HCPs) and Treatment Implementation, 3. Parent Factors (a. Beliefs and attitudes, b. Experience and impact of sleep problems, c. Knowledge), and 4. NDD-Specific factors.

Lack of knowledge about sleep, combined with limited availability of services and difficulty accessing available treatments, were frequently reported barriers by parents. When parents were able to access treatment, some reported negative experiences with HCPs such as feeling unheard or perceiving their HCPs as not knowledgeable about sleep and NDDs. For example, a parent commented, "I don't think there's a lot of information available to doctors around this. It seems to be an area that doesn't have a lot of research" (P32, ADHD). Some parents reported that HCPs only seemed to offer melatonin and medication as treatment options, and other parents expressed reluctance to use such treatments for sleep. A parent shared, "our doctors just automatically wanted to medicate [for sleep problems]" (P31, CP). Another parent stated, "The pediatrician who diagnosed my daughter with ADHD simply said as an aside, 'Oh for sleep, you know you can give her melatonin and you can do it long term,' and that was all that was ever said by him in the course of discussing her treatment" (P28, ADHD).

The negative impact of sleep problems on parents and families also acted as a barrier that influenced parents' decision to seek treatment and ability to implement treatment, as did their own feelings of self-blame, anxiety, and exhaustion. One parent felt that implementing strategies was difficult because, "we're kind of empty. We have no more gas left in the tank after five years of sleep deprivation" (P39, ADHD). Another parent said that it was hard to "be consistent with anything initially because you're just so tired that, even though you know what you should do, and you know what needs to be done […] you just do whatever you can to […] get them to bed, or get them to go back to bed in the night. It's kind of hard to be logical" (P20, ASD). Furthermore, addressing sleep problems was not always described as a priority; a parent shared, "My child's needs are so high and it's so intense all of the time […] we've never just made an appointment for sleep because we're really in the throes of the crises every day" (P29, FASD). For some parents, the complexity and comorbidity associated with their children's NDD diagnoses was reported to act as a barrier, especially NDD symptoms, NDD medications (especially stimulants and seizure medications), children's anxiety, attachment concerns, trauma history, and medical issues (e.g., seizures, feeding problems).

Facilitators. Three facilitators were identified, comprised of 24 individual codes (see Table 3.4): 1. Experience with Service (HCPs) and Treatment Implementation, 2. Parent Factors (a. Beliefs and attitudes, b. Education), and 3. Support. Overall, parents were able to identify some aspects of their experiences with HCPs and treatment that had facilitated their seeking or use of treatment: supportive and caring HCPs, a behavioural approach to treatment, and consistency with treatment implementation were particularly helpful. Some parents also reported that individualization of treatment (i.e., tailoring treatment to both child and parent needs) was helpful. Trying out different types of treatment was also reported to be helpful; one parent noted, "You just try different things, I guess. See what works" (P43, CP). Specific parental beliefs and attitudes were also reported to be facilitators, including being persistent, hopeful, self-advocating, and experiencing success. Parents reported self-education to be a facilitator, with some either doing their own research on sleep or drawing on their own specialized experience. One parent offered this perspective, “The books and the education and the establishing routines, those have all been quite helpful, or helpful to varying degrees. […] none of them have been perfect, but […] picking away at it from all directions has helped” (P15, ASD). Finally, support, especially from other parents, was identified as a key facilitator, with a parent sharing, “The parents are the people who help you the most. Because you learn from them. […] You learn not to give up” (P3b, ASD).

Differences across NDD groups. Most themes were common across all four NDD groups, and most differences were reported within the NDD-Specific Factors barrier. Some parents of children with FASD reported believing that sleep problems in their children were more complex to treat than in other NDDs, whether due to a history of trauma and attachment concerns, or because they perceived their children as less responsive to behavioural treatments due to neurological impairment. Parents of children with CP reported pain and medical problems (e.g., muscle tightness, limited mobility) as barriers to sleep more often than did parents of children with other NDDs; for example, some parents reported that pain appeared to cause their children’s sleep problems.

Health Care Professionals

Barriers. Five barriers were identified for HCPs, comprised of 32 individual codes (see Table 3.5): 1. Access to/Availability of Services, 2. HCP Factors (a. Education, training, and experience; b. Beliefs and attitudes), 3. Individual Practice Factors (a. Time, b. Supporting families, c. Nature of role/practice), 4. Parent Factors (a.

Parent ability to implement/follow through with treatment, b. Parents' access to treatment, c. Parental beliefs and attitudes), and 5. NDD-Specific Factors.

Health care professionals reported that both their own limited access to resources needed to provide sleep treatments, as well as their patients' limited access to sleep- and NDD-related services, could act as barriers to treatment provision. One HCP noted that a "lack of resources" about sleep and NDD meant there was "nowhere for parents to get […] help when they need it" (H34, CP). Some HCPs also highlighted their lack of experience and training with sleep as potential barriers, saying: "It doesn't really feel like we're experts in sleep… [because we] weren't trained through school to think about sleep as a targeted intervention or a targeted goal" (H8, ASD). Lack of time and availability to provide treatment and conduct follow-up appointments was another barrier. An HCP noted, "Continued support I think is the most important thing, but it's the hardest thing to do, given […] a clinic setting and availability of clinicians" (H11, ASD).

Health care professionals also identified parents' exhaustion, stress, and capacity for implementing treatment as barriers, noting that when children do not sleep, neither do their parents. An HCP indicated that if parents "don't identify [sleep] as a problem, then it's not really something I'm gonna tackle at that point for them," because treatment depended on parents "going through a pretty rough sleep to improve sleep behaviours" (H4, ASD). An HCP said, "It's not an easy fix and it's also not something that is fixed quickly, so that's difficult when parents are exhausted by the time they bring these problems to light […] their ability to cope is compromised from the get go" (H25, ADHD). Finally, some HCPs noted that specific NDD-related factors could be barriers to treatment, including comorbidities (mental health and medical) and use of medications that target NDD symptoms but may compromise sleep.

Facilitators. Three facilitators were identified, comprised of 22 individual codes (see Table 3.6): 1. HCP Factors (a. Education, training, and experience), 2. Individual Practice Factors (a. Supporting families, b. Collaboration), and 3.Treatment Approaches and Experience (a. Family-centered approach, b. Helpful treatment strategies, c. Modifications to treatment). In general, facilitators were related to HCPs' acquisition of knowledge and education about sleep problems in NDDs, working collaboratively with colleagues, their perceived ability to adequately support families, and a variety of specific approaches to treatment (including strategies and treatment modifications). Some HCPs reported that self-education was very helpful. Several mentioned using sleep/NDD resources such as the Autism Speaks Sleep Toolkit (https://www.autismspeaks.org/sleep). One HCP commented, "I don't have any formal training or education in [sleep]. It's more what I've learned through experience and what I've picked up in supporting families" (H2, ASD).

A family-centered treatment approach that incorporates parents' values and perspectives was recommended as facilitating provision. One HCP described treating sleep problems as "a partnership with parents" (H27, CP). Health care professionals reported that a behavioural approach to treatment could be a facilitator, particularly when psychoeducation about sleep was combined with the use of coaching and modelling strategies for parents. One HCP expressed that education was extremely important, saying, "Many families these days under-value sleep and under-appraise the importance of sleep and what the implications of lack of sleep are for children," and noting that the "number one" recommendation would be "educating families on how to better set up sleep hygiene and routines to accomplish that" (H38, ADHD). Finally, some HCPs noted that in addition to individualizing treatment to the child, addressing medical factors, adjusting children's NDD medication regimens (e.g., stimulants and anti-epileptics), and modifying treatments to accommodate NDD symptoms such as rigidity and other factors such as children's functional levels and feeding/swallowing problems were helpful.

Differences across NDD groups. Few differences across NDD groups were reported. Some HCPs suggested that more resources are available for sleep problems in the context of ASD than other NDDs. Similar to parents, a few differences emerged for FASD and CP. For example, some HCPs felt that sleep problems were harder to treat in children with FASD, because of the presence of dysregulation, brain damage, and history of trauma/attachment problems. Professionals working with children with CP also identified sleep problems as being primarily related to pain and medical factors, compared to the behavioural factors endorsed by the other HCPs.

Similarities and Differences Between Parent and Health Care Professionals.

Lack of information, awareness, and accessible services for sleep were reported to be barriers by both parents and HCPs. Parents and HCPs expressed concerns about each other, with some parents reporting that their experiences with HCPs could act as barriers or facilitators, and HCPs reporting concerns about not wanting to burden parents with unfeasible treatments or commenting on parents' inconsistent implementation. Both parents and HCPs acknowledged the difficulty of sleep treatment, emphasizing parental stress and exhaustion as potential barriers. Both parents and HCPs reported that in some cases, sleep problems were not prioritized for treatment amongst children's other behaviour problems (e.g., disruptive behaviours).

Knowledge and education were endorsed by both parents and HCPs as facilitators to treatment. Both also found the same treatment approaches helpful – primarily behavioural approaches, emphasis on consistency, use of bedtime routines, and healthy sleep habits, with incorporation of melatonin or medication as needed. Individualization of treatment also emerged as a theme amongst both parents and HCPs; for example, some parents reported needing to take an individualized, trial and error approach to treatment (i.e., trying out multiple treatments to find one that worked). From HCPs' perspectives, the ability to individualize and modify treatments to children's and parents' needs facilitated treatment provision (e.g., using more visual supports, addressing environmental sensitivities, accommodating functional level, adjusting time expectations). Similar core behavioural treatment strategies and modifications were identified as helpful across all four NDDs by parents and HCPs.

Discussion

The main purpose of this study was to identify barriers and facilitators experienced by parents and HCPs in accessing and utilizing treatment for sleep problems in children with NDDs, in order to better inform our understanding of treatment needs from both parents' and HCPs' perspectives, and to inform the development of a sleep intervention for children with NDDs. Key themes that emerged from the data were similar for both parents and HCPs. There is a general lack of knowledge and awareness about sleep problems among both parents and HCPs, combined with inaccessible or limited services and evidence-based treatments. Sleep problems and their treatment appear to be especially challenging, demanding, and intensive due to the negative impact on parents and the need to individualize treatment to children's needs within a complex array of NDD symptoms and comorbidities. Treatments often require already-tired parents to implement difficult strategies consistently night after night with tired, uncooperative children and little support from professionals. However, parents who had implemented sleep treatments and HCPs who provided sleep treatments for their patients with NDDs reported that perseverance with behavioural treatment, particularly consistent use of bedtime routines and healthy sleep habits, combined with melatonin or medication as needed, were effective and helpful. Given the intensity of sleep treatments, ensuring that families feel supported by their HCPs, motivated, and hopeful before beginning and throughout treatment is critical.

When the four NDD groups were compared, very few differences in barriers and facilitators emerged. The primary differences related to specific aspects of FASD and CP that could act as barriers to sleep treatment. However, across all NDD diagnoses, the same core behavioural strategies were reported to be used, with modifications to accommodate specific NDD symptoms. Although this transdiagnostic use of strategies initially appears to contradict the need for individualization of treatment, it should be noted that parents and HCPs understood individualization as tailoring treatment to a child's needs. The actual treatment approaches and specific strategies that they used and recommended were the same across all four disorders. This suggests that exploring a transdiagnostic approach to treatment may be useful, consistent with existing literature on sleep interventions for children with NDDs (Rigney et al., 2018).

Clinical Implications

Canadian parents of children with NDDs and HCPs working with these children reported that neither sleep treatments nor information and education about sleep are easily accessible. In particular, standard face-to-face treatment modalities may not be accessible or feasible, with HCPs sharing that they are not able to follow up adequately with parents. Online intervention delivery (i.e., eHealth) may offer a solution to these barriers, as it is more accessible and wider reaching than conventional face-to-face interventions (Breitenstein, Gross, & Christopherson, 2014). Another solution to reducing HCP time and involvement is parent-implemented interventions, wherein parents are trained to deliver treatments to their children directly. Such interventions have been shown to be effective for a wide range of NDD concerns (e.g., Althoff, Dammann, Hope, & Ausderau, 2019). Self-directed eHealth parent-implemented interventions may be an ideal vehicle for delivering sleep psychoeducation and behavioural strategies directly to parents. However, given the challenges that both parents and HCPs noted about being stressed and having difficulty following through with intervention implementation, it will be important to explore how to provide adequate support to parents.

Given emerging evidence that effective sleep treatment strategies are transdiagnostic across NDDs (Rigney et al., 2018), a modular transdiagnostic eHealth intervention likely has great potential (e.g., Sauer-Zavala et al., 2017). For example, such an intervention could offer general psychoeducation about sleep in the context of NDDs, and recommend core behavioural strategies, healthy sleep habits and bedtime routines (e.g., Rigney et al., 2018). If more specific NDD diagnostic information is required, parents could choose to access a module specifically about sleep in the context of their child's diagnosis.

The results of the present study have been used to inform the modification of the *Better Nights, Better Days* (BNBD) intervention for TD children with insomnia (Corkum et al., 2018) into *Better Nights, Better Days for Children with Neurodevelopmental Disorders* (BNBD-NDD). The original BNBD was recently the subject of a Canada-wide RCT (NCT02243501, clinicaltrials.gov). Based on the current research, along with the extant literature (see Rigney et al., 2018), BNBD-NDD was developed as a modular transdiagnostic parent-implemented eHealth intervention for parents of children with ASD, ADHD, CP, and FASD (see Tan-MacNeill et al., 2020, for results of usability testing).

Limitations

This sample of participants may have been more interested in or knowledgeable about sleep than other parents and HCPs, given their willingness to participate in an online study about sleep. Likewise, the study may have appealed to participants with greater internet literacy. Although we aimed to recruit a diverse and representative sample, parents of children with more severe sleep problems or other behavioural symptoms may have been less able to participate. Difficulties in scheduling participants necessitated the administration of interviews as well as focus groups, in order to accommodate participants. While emergent themes were consistent across interviews and focus groups during coding, nevertheless different information may have been gained from these two approaches. The themes that emerged from the data may also have been influenced by the questions asked in the topic guides. Finally, the study was expanded from originally only including ASD-specific participants to include the other three NDDs to inform the development of the BNBD-NDD intervention. As such, ASD-specific participants are overrepresented in the sample and recruitment of groups was non-concurrent (but all completed within a two-year window).

Conclusion

Overall, these findings suggest a great need for more awareness about the importance of healthy sleep for children with NDDs, more education about how to treat sleep problems, and more evidence-based interventions that are readily accessible. Similar barriers, facilitators, and effective treatment strategies were identified across all four NDDs, suggesting that a transdiagnostic approach to treatment would be helpful. An eHealth intervention would address many of the reported barriers to treatment.

Table 3.1

Demographic Information for Parent Participants and their Children

	Total	Primary NDD Group			
		ASD	ADHD	CP	FASD
	N = 43	(*n* = 20)	(*n* = 9)	(*n* = 6)	(*n* = 8)
Parent Participant Demographics					
Participants' relationship to child					
Biological Mother	32 (74.4%)	18 (90%)	9 (100%)	5 (83.3%)	
Biological Father	3 (7%)	2 (10%)		1 (16.7%)	
Adoptive Mother	8 (18.6%)				8 (100%)
Participant (Parent/caregiver) Mean Age in Years (SD, range)	38.5 (SD = 7.1, 25-65)	36.5 (SD = 5.2, 25-47)	38.2 (SD = 4.7, 32-45)	38.7 (SD = 8.5, 28-52)	43.6 (SD = 10.5, 32-65)
Participants' relationship status					
Married / Common-law	33 (75.7%)	15 (75%)	6 (66.7%)	5 (83.3%)	7 (87.5%)
Single / Never legally married	3 (7%)	1 (5%)		1 (16.7%)	1 (12.5%)
Separated / Divorced	6 (14.0%)	4 (20%)	2 (22.2%)	1 (16.7%)	1 (12.5%)
Community of residence					
Rural	12 (27.9%)	5 (25%)	2 (22.2%)	1 (16.7%)	4 (50%)
Town	5 (11.6%)	3 (15%)	1 (11.1%)	1 (16.7%)	1 (12.5%)
City	26 (60.4%)	12 (60%)	6 (66.7%)	4 (66.7%)	3 (37.5%)
Ethnic or Cultural Heritage					
White / Caucasian	39 (90.7%)	19 (95%)	8 (88.9%)	6 (100%)	6 (75%)
Aboriginal – Metis	1 (2.3%)				1 (12.5%)
Other	2 (4.7%)	1 (5%)			1 (12.5%)
Highest Level of Education					
High school equivalent or less	3 (7%)	2 (10%)		1 (16.7%)	
Diploma or certificate from college, university,	18 (41.2%)	7 (35%)	5 (55.5%)	2 (33.3%)	4 (50%)

	Total	Primary NDD Group			
	N = 43	ASD (*n* = 20)	ADHD (*n* = 9)	CP (*n* = 6)	FASD (*n* = 8)
trade/technical/vocational school, or less					
Bachelor’s/ Undergraduate Degree (e.g., BA, BSc, BEd)	13 (30.2%)	7 (35%)	2 (22.2%)	2 (33.3%)	2 (25%)
Graduate degree (e.g., MA, MSc, MEd, PhD, DSc, EdD)	8 (18.6%)	4 (20%)	1 (11.1%)	1 (16.7%)	2 (25%)
Participant’s Current Employment Status					
Full Time	21 (48.8%)	7 (35%)	6 (66.7%)	4 (66.7%)	4 (50%)
Part Time	7 (16.3%)	6 (30%)			1 (12.5%)
Unemployed	1 (2.3%)				1 (12.5%)
Student	1 (2.3%)	1 (5%)			
Homemaker	8 (18.6%)	6 (30%)		2 (33.3%)	
Other	4 (9.3%)		2 (22.2%)		2 (25%)
Estimated Household Income					
Less than $30,000	3 (7%)	2 (10%)		1 (16.7%)	
$30,000 - $59,999	7 (16.3%)	3 (15%)	1 (11.1%)	1 (16.7%)	2 (25%)
$60,000 - $99,999	16 (37.2%)	6 (30%)	4 (44.4%)	1 (16.7%)	3 (37.5%)
$100,000 +	16 (37.2%)	9 (45%)	2 (22.2%)	2 (33.3%)	3 (37.5%)
$100,000 - $149,999	9 (20.9%)	6 (30%)	1 (11.1%)	2 (33.3%)	
$150,000 - $199,999	5 (11.6%)	1 (5%)	1 (11.1%)		3 (37.5%)
$200,000 and over	2 (4.7%)	2 (10%)			
Average Number of Other Children in Home (mean; SD; range)	1.95 (.90, 1-5)	1.85 (.49; 1-3)	2.44 (1.13, 1-5)	1.50 (.55, 1-2)	2.00 (1.41, 1-5)
Child Demographics					
Child Sex					
Male	29 (67.4%)	15 (75%)	6 (66.7%)	3 (50%)	5 (62.5%)
Female	14 (32.6%)	5 (25%)	3 (33.3%)	3 (50%)	3 (37.5%)

	Total	Primary NDD Group			
	N = 43	ASD (*n* = 20)	ADHD (*n* = 9)	CP (*n* = 6)	FASD (*n* = 8)
Child Mean Age in years (SD, range)	8.5 years (SD = 2.5, 4.3 – 12.6)	9 years (SD = 2.5, 4.8 – 12.6)	6.9 years (SD = 2.3, 4.3 – 11.4)	8.3 years (SD = 2.3, 4.9 – 11.7)	9 years (SD = 2.3, 4.8 – 11.5)
Years Since NDD Diagnosis (SD, range)	4.3 (2.5, 1-10)	4.1 (SD = 1.8, 2-8)	2.6 (SD = 2.3, 1-7)	5.8 (SD = 1.9, 4-9)	5 (SD = 3.6, 1-10)
Comorbid Diagnoses: Additional NDD, Mental Health, and Physical Disorders (may have multiple diagnoses)					
Presence of Parent-reported Diagnosis	30 (69.8%)	12 (60%)	5 (55.6%)	5 (83.3%)	8 (100%)
Another NDD (ADHD, ASD, CP, or FASD)	9 (20.1%)	2 (10%)		1 (16.7%)	6 (75%)
Learning Disability	7 (16.3%)	2 (10%)	1 (11.1%)	2 (33.3%)	2 (25%)
Intellectual Disability / Developmental Delay	9 (20.9%)	3 (15%)		2 (33.3%)	4 (50%)
Mental Health disorder (e.g., at least one of: anxiety, mood, obsessive compulsive disorder, oppositional defiant disorder)	26 (60.5%)	10 (50%)	5 (55.6%)	4 (66.7%)	7 (87.5%)

Note: One participant did not complete; multiple participants missed or skipped questions.

Table 3.2

Demographic Information for Health Care Professionals

	Total	Primary NDD Group			
	(*N* = 44)	ASD (*n* = 21)	ADHD (*n* = 8)	CP (*n* = 8)	FASD (*n* = 7)
HCP Sex					
Male	3 (7%)	1 (4.8%)	0	0	2 (28.6)
Female	41 (93%)	20 (95.2%)	8 (100%)	8 (100%)	5 (71.4%)
Highest Level of Education					
Bachelor's Degree	9 (20.9%)	3 (14.3%)	2 (25%)	3 (37.5%)	1 (14.3%)
Master's Degree	18 (41.9%)	12 (47.1%)	1 (12.5%)	3 (37.5%)	2 (28.6%)
MD	5 (11.6%)	2 (9.5%)	2 (25%)		1 (14.3%)
PhD	8 (18.6%)	3 (14.3%)	2 (25%)	1 (12.5%)	3 (42.9%)
Years of Practice (Mean, SD, range)	14.5 (SD = 10.9, 0.5 – 38)	12.7 (SD = 10.8, 1-33)	13.9 (SD = 9.2, 1.5 – 31)	19.0 (SD = 13.8, 2-38)	15.4 (SD = 9.8, 0.5 – 30)
Practice Area					
Primarily health	27 (61.4%)	12 (57.1%)	5 (62.5%)	7 (87.5%)	3 (42.9%)
Primarily mental health	9 (20.5%)	7 (33.3%)	1 (12.5%)		1 (14.3%)
Evenly split between health / mental health	3 (6.8%)	1 (4.8%)			2 (28.6%)
Practice Setting					
Private practice	9 (20.5%)	3 (14.3%)	3 (37.5%)		3 (42.9%)
Community health or mental health centre	7 (15.9%)	5 (23.8%)			2 (28.6%)
Hospital	14 (31.8%)	11 (52.4%)	1 (12.5%)	2 (25%)	
University	1 (2.3%)		1 (12.5%)		
Other (e.g. non-profit, rehab, treatment centre)	8 (18.2%)	1 (4.8%)	1 (12.5%)	5 (62.5%)	1 (14.3%)

Years of Experience Working with Children with NDDs (Mean, SD, range)	13.9 (SD = 9.9, 2-35)	10.5 (SD = 6.5, 2-30)	15.0 (SD = 11.7, 2-30)	16.2 (SD = 31.1, 2.5 - 33)	20.3 (SD = 10.9, 8 – 35)
Self-reported estimated percentage of practice time working with children with NDD (Mean, SD, range)	48.6% (SD = 31.1, 0.2 – 100)	59.7% (SD = 28.6, 3 – 100)	25.4% (SD = 30.3, 0.2 – 75)	38.6% (SD = 22.5, 5-70)	47.0% (SD = 36.7, 5-90)
Specialize in NDDs?					
Yes	34 (77.3%)	19 (90.5%)	3(37.5%)	6 (75%)	6 (85.7%)
No	10 (22.7%)	2 (9.5%)	5 (62.5%)	2 (25%)	1 (14.3%)

Note: Three participants did not complete and one only partially completed the demographic / background information questionnaires.

Table 3.3

Parent Barriers and Codes

Barrier	Codes
1. Access to / Availability of Services	
	1. Long wait times
	2. NDD specialists difficult to access or not available
	3. Need to access multiple HCPs or disciplines
	4. Not able to attend appointments
	5. Sleep treatment not affordable
	6. Lack of available information & resources
2. Experience with Service (HCPs) and Treatment Implementation	
	7. HCPs lack knowledge about sleep & NDDs
	8. Perceptions of HCPs as not helpful
	9. Negative interpersonal experience with HCPs
	10. Behavioural treatment can lead to a behaviour burst or dysregulation (unwanted)
	11. Inconsistent response to treatment
	12. Individualization – no one size fits all treatment
	13. Treatment not working
	14. Trial and error (don't know what will work / why)
	15. Treatment is hard (challenging)
3. Parent Factors	
3A. Beliefs & Attitudes	
	16. Reluctance to stop using what works even if problematic (e.g., co-sleeping)
	17. Reluctance to use medication for sleep
	18. Reluctance to use melatonin for sleep
	19. Cultural beliefs – co-sleeping acceptable
	20. Expectation of negative outcome
	21. Belief that child's brain is wired differently in NDD
	22. Belief that sleep tips for TD don't apply for NDDs
3B. Experience and Impact of Sleep Problems	
	23. Parental guilt / self-blame / anxiety for sleep problem
	24. Feeling judged /stigmatized by others
	25. Sleep is not first priority
	26. Caregivers have different perspectives about sleep
	27. Negative impact on family
	28. Parental exhaustion & stress

Barrier	Codes
3C. Knowledge	
	29. Lack of awareness about sleep in NDDs
	30. Lack of knowledge of underlying cause of sleep problem
	31. Lack of knowledge of where to go for help or what to ask
4. NDD-Specific Factors	
	32. Complexity and comorbidity associated with NDDs complicates sleep treatment (e.g., child anxiety, attachment concerns, trauma history, physiological issues)
	33. NDD medications negatively affect sleep
	34. NDD symptoms make sleep problems harder to treat (e.g., needing to wind down; limited communication ability; pain / physical symptoms in CP; difficulty taking medication; level of functioning; rigidity / difficulty with transitions; sensory sensitivities)

Table 3.4

Parent Facilitators and Codes

Facilitator	Codes
1. Experience with Service (HCPs) and Treatment Implementation	
	1. Supportive, caring HCPs
	2. Behavioural approach to treatment
	3. Consistency
	4. Incorporating medication
	5. Incorporating melatonin
	6. Incorporating sensory or physiological components
	7. Individualization of treatment to child's needs
	8. Involving child in treatment
	9. Nutrition
	10. Practicing healthy sleep habits
	11. Same strategies work for TD
	12. Trying out different treatments
	13. Understanding what's comfortable for both child and parent
	14. Using bedtime routines
2. Parent Factors	
2A. Beliefs and Attitudes	
	15. Hope or past experience of success
	16. Persistence or keeping going
	17. Willing to try anything
	18. Self-advocacy
2B. Education	
	19. Discovering cause of sleep problem (e.g., by assessment)
	20. Drawing on own specialized experience
	21. Getting psychoeducation about sleep
	22. Self-education & doing own research
3. Support	
	23. Having support
	24. Support from other parents

Table 3.5

Health Care Professional Barriers and Codes

Barrier	Codes
1. Access to / Availability of Services	1. Lack of / limited specialist evidence-based sleep treatment & NDD services
	2. Lack of information and resources
2. HCP Factors 2A. Education, Training & Experience	3. Lack of experience or training with sleep
	4. Limited awareness of importance of sleep
	5. Perceived self-efficacy – not sleep experts
2B. Beliefs & Attitudes	6. Different approaches from different HCPs
	7. Relying on anecdotal data rather than functional behaviour analysis
	8. Some strategies work, some don't (hit or miss)
3. Individual Practice Factors 3A. Time	9. Lack of time and availability to provide treatment
	10. Lack of time to access information and educate self
3B. Supporting Families	11. Unable to provide adequate or direct support
3C. Nature of Role/Practice	12. Nature of role / service = limited involvement or capacity for sleep treatment
	13. Outside scope of practice
4. Parent Factors 4A. Parent Ability to Implement & Follow Through with Treatment	14. Caregivers lack support
	15. Challenging to get parents to implement strategies / follow through consistently
	16. Concern that parents do not have capacity to implement treatment (treatment not feasible)
	17. Lack of stable home environment
	18. Parental mental health concerns
	19. Parents are exhausted / stressed / burned out

Barrier	Codes
4B. Parents’ Access to Treatment	
	20. Language & communication are treatment barriers
	21. Parents not able to physically attend appointments
	22. Treatments not affordable / cost too great
4C. Parent Beliefs & Attitudes	
	23. Cultural norms conflict with recommended behavioural strategies (e.g., co-sleeping)
	24. Parents not ready for treatment
	25. Parents don’t know that sleep problems can be treated/think they are normal
	26. Medications are preferred/more frequently used
	27. Parents are concerned about/resistant to using medication/melatonin
	28. Parents are desperate for immediate solution
	29. Sleep is not parents’ main priority for treatment
5. NDD-Specific Factors	
	30. Complexity and comorbidity associated with NDD complicates sleep treatment
	31. Medication for NDD symptoms negatively impacts sleep
	32. NDD symptoms make sleep problems harder to treat

Table 3.6

Health Care Professional Facilitators and Codes

Facilitator	Codes
1. HCP Factors	
1A. Education, Training, & Experience	1. Professional development or formal training in sleep 2. Self-education 3. Accessing evidence-based literature 4. Accessing & using pre-existing resources
2. Individual Practice Factors	
2A. Supporting Families	5. Ability to provide direct support to families 6. Ability to work in-home (e.g., BCBAs)
2B. Collaboration	7. Consultation with other colleagues 8. Multidisciplinary team approach
3. Treatment Approaches and Experience	
3A. Family-Centered Approach	9. Accommodating and understanding that caregivers may be on different pages 10. Making treatment manageable for parents and preparing them for difficulties 11. Taking into account family values and parents' perspective and understanding of sleep 12. Help families experience success & positive affirmation
3B. Helpful Treatment Strategies	13. Behavioural approach to treatment 14. Consistency (helping families maintain) 15. Generalization of strategies across diagnoses 16. Psychoeducation about sleep to parents 17. Using assessment to inform sleep treatment 18. Using coaching, modelling, and teaching of strategies to parents
3C. Modifications to Treatment	19. Addressing physiological or physical factors affecting sleep 20. Individualization of treatment to the child 21. Modifying NDD medication regimen

Facilitator	Codes
	22. Modifying treatments for NDD symptoms is helpful (e.g., accommodating functional level, adapting strategies for NDD severity, addressing rigidity / difficulty with transitions, helping parents adjust expectations, addressing feeding / swallowing issues, focusing on routines, modifying environment, accommodating sensory sensitivities, using visual supports)

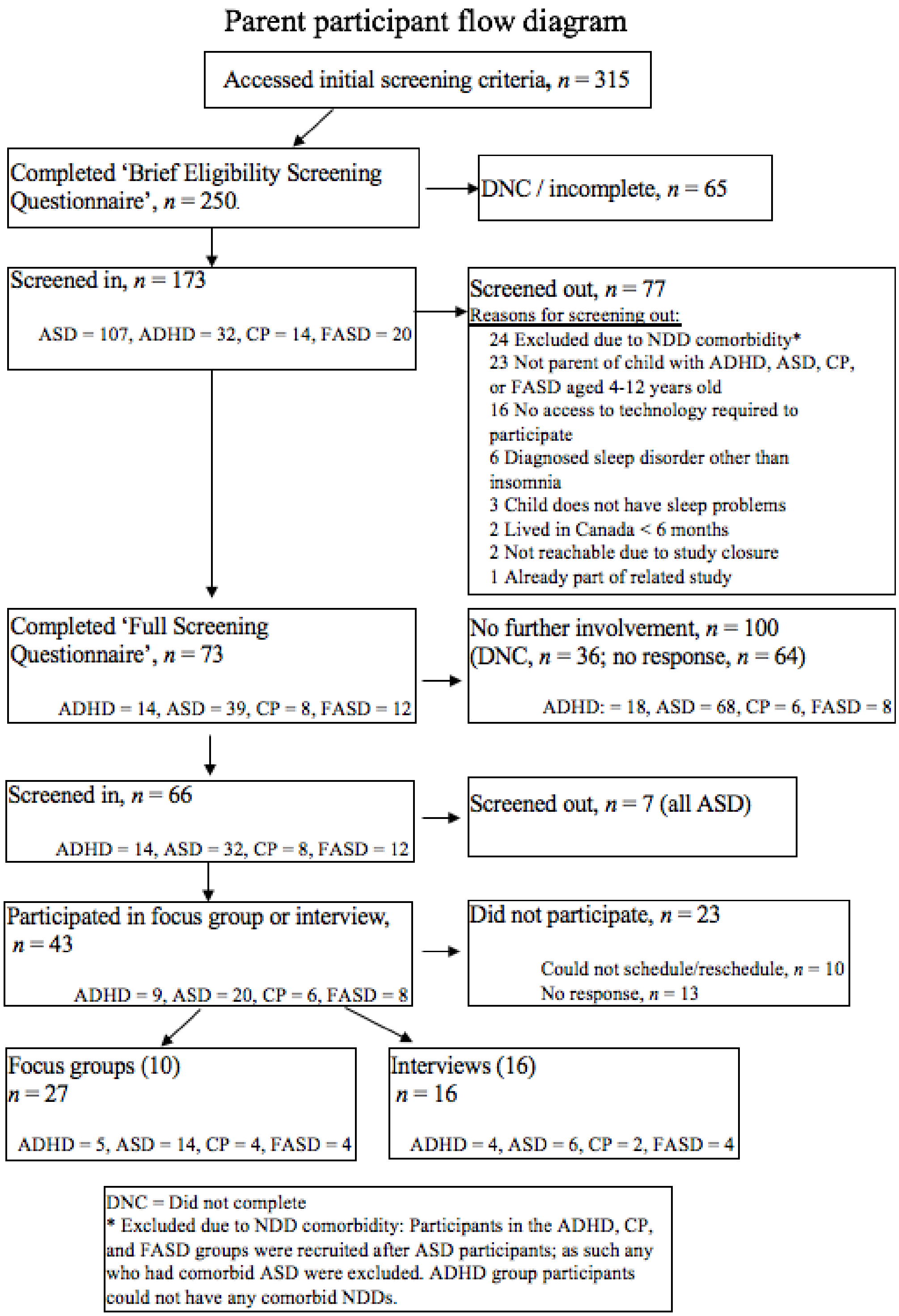

Figure 3.1. *Parent Study Flow Diagram*.

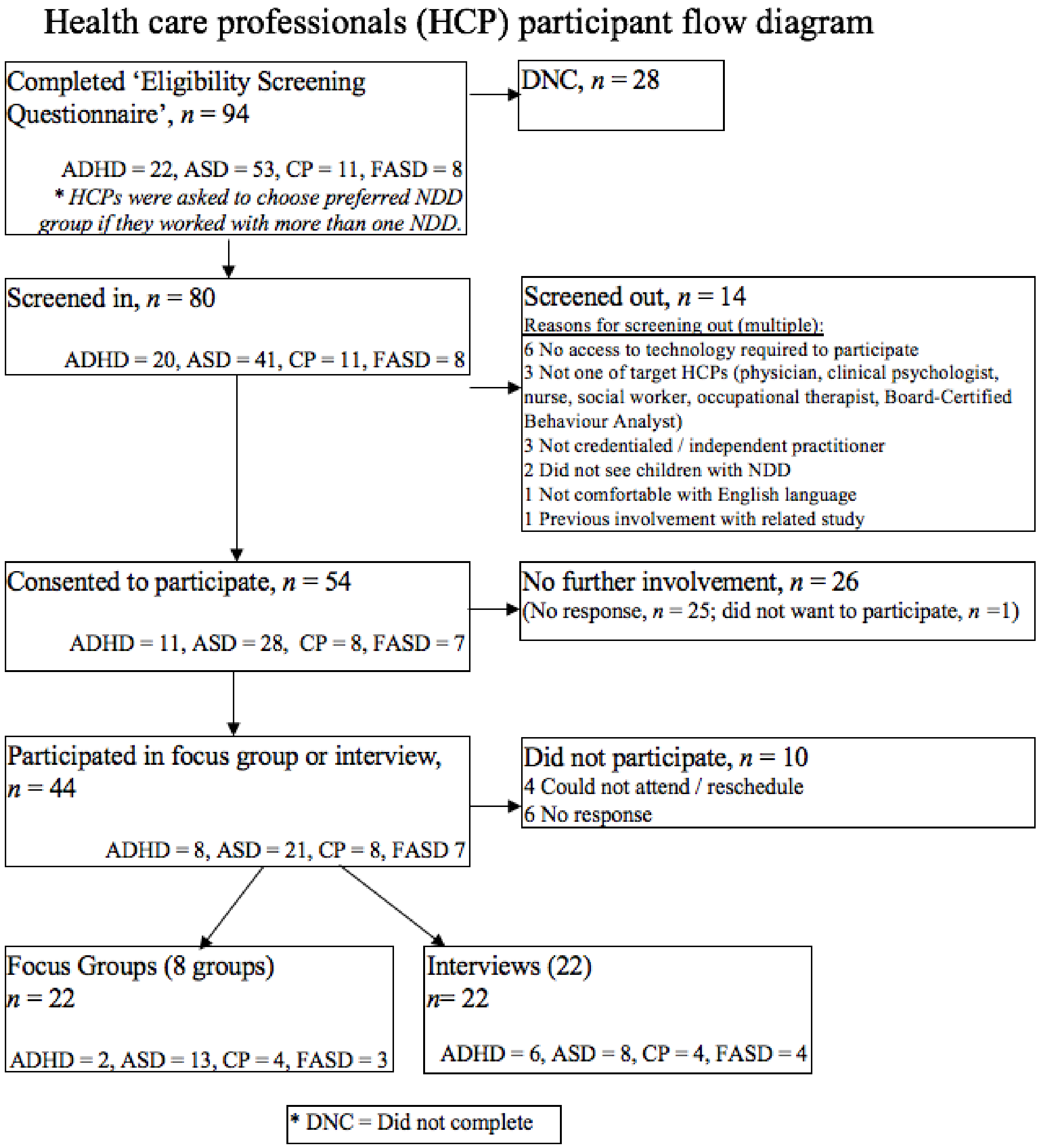

Figure 3.2 HCP Study Flow Diagram.

Chapter 4: An eHealth Insomnia Intervention for Children with Neurodevelopmental Disorders: Results of a Usability Study

The manuscript based on this experimental study is presented below. Readers are advised that Kim M. Tan-MacNeill, under the supervision of Dr. Isabel Smith and Dr. Penny Corkum, was responsible for developing the research questions, conducting the background literature review, applying for and obtaining research ethics approval, recruiting participants, completing and overseeing data collection, data analysis / interpretation, and all aspects of the writing process. All aspects of this research were done in consultation with Dr. Jill Chorney and Dr. Shannon Johnson, dissertation committee members. The BNBD-NDD research team also provided editorial feedback, including Dr. Shelly Weiss, Dr. Evelyn Constantin, Dr. Sarah Shea, Dr. Ana Hanlon-Dearman, Dr. Cary A. Brown, Dr. Roger Godbout, Dr. Osman Ipsiroglu, and Dr. Graham J. Reid. Data were collected and managed using the RedCAP database, an electronic data capture tool (Harris et al., 2009; Harris et al., 2019).

This manuscript was published as:

Tan-MacNeill, K.M., Smith, I.M., Weiss, S.K., Johnson, S.A., Chorney, J., Constantin, E., Shea, S., Hanlon-Dearman, A., Brown, C.A., Godbout, R., Ipsiroglu, O., Reid, G. J., & Corkum, P.V. (2020). An eHealth insomnia intervention for children with neurodevelopmental disorders: Results of a usability study. *Research in Developmental Disabilities, 98*, 1-14. doi: https://doi.org/10.1016/j.ridd.2020.103573

Abstract

Background: Sleep problems, particularly insomnia, are highly prevalent in children with neurodevelopmental disorders (NDDs) and can negatively affect health and development. eHealth interventions may increase access to evidence-based care for insomnia for children with NDDs, as programs are rare in most communities. *Better Nights, Better Days* (BNBD) is an online, parent-implemented intervention for pediatric insomnia in typically developing 1- to 10-year-olds.

Aims: The present study examined whether parents of children with NDDs perceived the original BNBD to be usable, acceptable, and feasible, and what modifications might be necessary to adapt it for children with NDDs.

Methods and Procedures: Twenty Canadian parents/caregivers of children aged 4-10 years with NDDs and insomnia implemented the BNBD intervention with their children, and completed usability questionnaires. Questionnaire data were analyzed quantitatively (descriptive statistics) and qualitatively (thematic analysis).

Outcomes and Results: Participants reported the intervention to be usable, useful, acceptable, and feasible. Several modifications were suggested to make the intervention more appropriate and acceptable for use with children with NDDs.

Conclusions and Implications: Results support a largely transdiagnostic approach to treating sleep in children with NDDs, and will inform the development of BNBD for Children with Neurodevelopmental Disorders (BNBD-NDD).

Introduction

Sleep Problems in Children with Neurodevelopmental Disorders (NDD)

Neurodevelopmental disorders (NDDs) emerge early in development and are characterized by differences that result in functional impairments in personal, social, academic and occupational domains (American Psychiatric Association, 2013). Between 50 and 90% of children with NDDs experience sleep problems (Cortese, Faraone, Konofal, & Lecendreux, 2009; Didden & Sigafoos, 2001; Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008; Robinson-Shelton & Malow, 2016; Romeo et al, 2014; ; Stade et al., 2008); insomnia, defined as difficulty falling or staying asleep, and/or night and early morning awakenings (Moore, Meltzer, & Mindell, 2007), is most common (Bruni et al., 2018; Richdale & Schreck, 2009). Physiological factors, child-parent interactions, environment, and NDD symptoms (e.g., inflexibility / rigidity, sensory sensitivities, self-regulation difficulty, anxiety) are all thought to contribute to the development and maintenance of insomnia in these children (Jan et al., 2008; Jan et al., 2010; Jan, Bax, Owens, Ipsiroglu, & Wasdell, 2012; Reynolds & Malow, 2011).

Sleep problems negatively affect the daytime functioning of children with NDDs, including increased severity of NDD symptoms (Tudor, Hoffman, & Sweeney, 2012) and emotional and behavioural problems (Goldman, McGrew, Johnson, Richdale, Clemons, & Malow, 2011), and potential interference with the effectiveness of interventions for the NDD (Vriend, Corkum, Moon, & Smith, 2011). Parents of children with NDDs believe that their children's sleep problems are less treatable than those of TD children (Bessey, Coulombe, Smith, & Corkum, 2013), reporting that they do not know what to do and are exhausted themselves (Ipsiroglu, McKellin, Carey, & Loock, 2013).

Sleep Treatments for Children with NDDs

Behavioural interventions are the first-line treatment for pediatric insomnia for both children with NDDs and TD children (Meltzer & Mindell, 2014; Malow et al., 2012). Research on behavioural interventions for sleep problems in children with NDDs includes several recent randomized controlled trials (RCTs) (e.g., Adkins et al., 2012; Corkum et al., 2016). A systematic review of parent-delivered behavioural sleep interventions for children with NDDs found support for a transdiagnostic approach to treating sleep problems (i.e., applying the same treatment principles across multiple NDD diagnoses), identifying psychoeducation, healthy sleep practices, graduated extinction and reinforcement as the most common interventions, with minor or no modification of strategies for TD children (Rigney et al., 2018).

Families of children with NDDs already experience barriers to evidence-based health care, including limited access to NDD specialists and long waits (Johnson & DeLeon, 2016), so access to intervention for sleep problems is an important consideration. Online or eHealth interventions delivered through the internet hold enormous potential for reducing barriers and increasing access. To our knowledge, an eHealth intervention for sleep problems in children with NDDs does not yet exist. Reviews of internet-based interventions designed to be delivered by parents for a broad range of clinical concerns suggest that such programs are effective for both TD children (Hall & Bierman, 2015) and children with NDDs (Meadan & Daczewitz, 2015).

Online interventions may create behaviour change through interactions among the user (e.g., a parent), website, and the environment (Ritterband, Thorndike, Cox, Kovatchev, & Gonder-Frederick, 2009). Given that website characteristics and construction impact intervention effectiveness, usability testing is a critical part of developing an online intervention and assessing its readiness for use. The *user experience honeycomb* is a frequently used framework for usability evaluation that breaks usability into seven inter-related facets of user experience: *usable, findable, useful, credible, desirable, accessible*, and at the centre, *valuable* (Morville & Sullenger, 2010).

Development and Usability Testing of *Better Nights, Better Days for Children with Neurodevelopmental Disorders (BNBD-NDD)*

Given the need for an accessible insomnia intervention for children with NDDs, and because sleep problems are common across NDDs, interest is great in developing a transdiagnostic intervention for use across diagnoses (Harvey, 2008) and delivered by parents. A modular treatment, wherein parents select evidence-based treatment strategies according to their needs, could be used for a range of diagnoses, in contrast with a one-size-fits-all treatment (Sauer-Zavala et al., 2017).

Emerging evidence suggests that the same behavioural strategies that are effective for treating pediatric insomnia in TD children may help those with NDDs, and that a transdiagnostic approach may be effective across NDD diagnoses (Rigney et al., 2018; Ali et al., 2018). Corkum and colleagues developed *Better Nights, Better Days* (BNBD), a parent-implemented eHealth intervention for use with 1- to 10-year-old TD children with insomnia (see Corkum et al., 2018 and Speth et al., 2015). A Canada-wide RCT has just been completed [NCT02243501, clinicaltrials.gov]. With the aim of modifying BNBD into BNBD-NDD, three studies have been conducted: 1) a systematic review of parent-implemented behavioural interventions for pediatric sleep problems in NDDs

(Rigney et al., 2018), 2) a qualitative exploration of barriers and facilitators to sleep treatment access / provision from NDD caregivers' and health care professionals' perspectives (Tan-MacNeill, Jemcov, Smith, & Corkum, 2017), and 3) a Delphi study to elicit recommended intervention content from health care professionals (Ali et al., 2018).

The present usability study is the final step of this process, preceding an efficacy trial. To explore whether a transdiagnostic intervention is appropriate for treating sleep problems in children with NDDs, we included four diverse and prevalent NDDs in this study: attention-deficit / hyperactivity disorder (ADHD), autism spectrum disorder (ASD), cerebral palsy (CP), and fetal alcohol spectrum disorder (FASD). Parents / caregivers of children with NDDs were presented with *the original, unmodified* BNBD program, and fully implemented the intervention with their children. They were asked about their experiences, including whether they had suggestions to make the program more usable, feasible, and acceptable for children with NDDs. Usability was defined as consisting of the user experience categories conceptualized in Morville's *user experience honeycomb* (Morville & Sullenger, 2010); feasibility referred to participants being able to fully participate in the intervention (e.g., complete activities, use strategies) without major obstacles; and acceptability pertained to participants finding the intervention acceptable as parents of children with NDDs (after using the original program for TD children).

Research Question

The primary research question was: Do parents/caregivers of children with NDDs perceive the unmodified BNBD intervention to be usable, acceptable, and feasible, and what modifications do they suggest to make it more so? Although a growing body of research suggests that transdiagnostic approaches to treating insomnia are effective with minimal modifications (e.g., Rigney et al., 2018), parents of children with NDDs report a preference for treatment tailored to their children's needs (Tan-MacNeill et al., 2017). Given this, we expected that participant feedback would indicate that the BNBD intervention is generally usable and feasible, but may require some diagnosis-specific modifications to be more acceptable to parents of children with NDDs.

Method

Participants

This study was approved by the Research Ethics Board of the IWK Health Centre in Halifax, Nova Scotia, Canada. Informed consent was obtained from all participants, who were primarily recruited online via social media.

The final sample included 20 Canadian parents or caregivers (hereafter, parents) of children aged 4 – 10 years with formal diagnoses of ADHD, ASD, CP and/or FASD from a physician or psychologist, as reported by parents. Inclusion and exclusion criteria are shown in Table 4.1. Children's ages were restricted to 4 – 10 years, as some NDDs cannot be reliably diagnosed in younger children. Comorbidities were present among the diagnoses (see Table 4.1 for primary / comorbid disorder criteria for group membership). The study sample included parents whose children had a range of disorders and difficulties. Detailed demographic information, including comorbid diagnoses, is reported in Table 4.2. Overall, the sample was largely Caucasian (80%), well-educated (all had some post-secondary education), English-speaking (90%), primarily mothers, and had a mean age of 41.9 years (range 29 - 57 years). The majority of children were male (85%) with a mean age of 8.99 years (SD = 1.57; range 5.10 - 10.90 years).

Initially, 27 parents were enrolled. One participant was excluded from analyses due to misunderstanding the study's purpose, as she believed the intervention to be already adapted for NDDs. Please see Figure 4.1 for a study flow diagram that depicts session and Usability Questionnaire completion rates and withdrawals. Fifteen parents completed the entire study. Data were analyzed for all parents who filled out at least one usability questionnaire (ranging from 20 participants in Session 1 to 15 in Session 5). Parent completion rates by session are shown in Table 4.3. Given the small sample and that the study purpose was to explore whether parents of children with a range of disability severity found the intervention to be useful, we did not examine differences between those who did and did not complete the intervention.

Parent-reported child sleep problems are described in Table 4.4. Children had to meet criteria for behavioural insomnia (Anders & Dahl, 2007) to be eligible (described in Measures). All parents endorsed children's problems falling asleep; 65% also had problems following bedtime routines, and 45% needed adults present to fall asleep. Thirty-five percent of parents were currently using melatonin to treat their children's sleep problems. Sixty-five percent of parents reported preferring to sleep separately from their children, while 30% preferred to sleep separately but co-slept because their children did not sleep well alone, and one parent reported co-sleeping due to space limitations. To take part in the study, parents were required to sleep separately from their children for the study duration, as the behavioural intervention focused on teaching children independent self-soothing skills at nighttime. Those who endorsed preferring to continue co-sleeping were not eligible to participate; one participant withdrew from the study, choosing to resume co-sleeping.

Description of Intervention

Parents were given online access to the original BNBD intervention, a five-session, parent-directed eHealth program for treating behavioural insomnia in TD children between ages 1 and 10 years (Corkum et al., 2018). Parents watched a “How to Use” tutorial prior to beginning. Intervention content included: Session 1 – general psychoeducation about children’s sleep and sleep problems; Session 2 – healthy sleep practices, bedtime routines, and nap scheduling; Session 3 – strategies for independent settling to sleep at bedtime; Session 4 – strategies for night waking, napping, and early morning awakenings; Session 5 – maintenance and preparing for future. Each session was designed to take 1-2 hours to complete online, involving videos, reading, activities, and interactive tools (e.g., building a sleep routine); participants could pause and resume sessions. All session activities / homework were designed to be completed within two weeks. Before progressing to the next session, parents needed to complete at least 5 daily sleep diaries (within a period of 14 days) for their children and wait at least seven days to allow time to try the session strategies. For more information, see Corkum et al. (2018) or visit http://betternightsbetterdays.ca.

Measures

Screening and Eligibility. Screening was conducted online in two steps:

1) Parents completed a 26-item, author-made questionnaire that asked broad questions to quickly target the inclusion and exclusion criteria (see Table 4.1) and minimize respondent burden on potential participants (see Appendix H for questionnaire).

2) If initial inclusion/exclusion criteria were met, parents completed the Behavioural Insomnia Questionnaire (BIQ; Anders & Dahl, 2007; modified by authors), Pediatric Sleep Questionnaire (PSQ; Chervin, Hedger, Dillon, & Pituch, 2000), Health-Related Questionnaire-Modified (HRQ-M; author-made – see Appendix I), and the Single Item Literacy Scale (SILS; Morris, MacLean, Chew, & Littenberg, 2006). The BIQ provides a cut-off score to determine presence of sleep onset (e.g., falling asleep, settling) or night-waking problems over the past month; authors' additions were parents' ratings of the perceived severity of children's sleep problems, impact on child and family functioning across domains (e.g., fatigue, mood, school, family life), and reports of co-sleeping.

Background Information. Eligible parents completed a Demographic Information Questionnaire (author-made; based on Canadian census format) and Treatment Utilization Questionnaire (Reid, 2005; history of treatment seeking and use for sleep and other health problems).

Usability Questionnaires. Parents completed an online End of Session Usability Questionnaire (ES-UQ; 31 items; author-made; Appendix J) after each of five sessions, and an Overall Usability Questionnaire (O-UQ; 62 items; author-made; Appendix K) at the end of the intervention. Parents rated their agreement with statements about the intervention's usability, usefulness, findability, desirability, credibility, accessibility, value, acceptability for NDDs, feasibility of implementation, readiness for use with children with NDDs, and perceived support (i.e., whether they felt supported in their use of the intervention or felt that they required more intensive/interactive support to utilize the website). Parents were also asked about their perceptions of the intervention activities (i.e., strategies they learned to implement with their children), videos, homework, and other features (such as printable summaries and worksheets). Ratings were on a 5-point Likert scale (e.g., 1 = strongly agree, 5 = strongly disagree), such that lower numerical ratings indicated greater agreement that the intervention or session was usable, useful, etc. Comments were optional (e.g., "Please provide comments that support your ratings about the ____ of this session. Include any suggestions you may have to improve ___."). These questionnaires are available upon request; UQ items and areas questioned are in results tables, below.

Usability Testing

After screening, eligible parents were enrolled in the study, given secure access to the BNBD intervention, and asked to implement it (see Appendix L for "How to Manual for Parents"). After each session, they were asked to provide feedback via the ES-UQs about whether the session was usable, feasible and acceptable, or how it could be modified for parents of children with NDDs. Parents were emailed links to the ES-UQs one week after completing each session to allow for implementation of strategies taught. After completing all five sessions and the corresponding ES-UQs, parents completed the O-UQ, and participated in an exit interview (with author K.T.M.). Exit interview data will be described elsewhere. Parents received up to a $100 gift card ($20 per session).

Analysis

Quantitative analyses. Descriptive statistics were calculated for participant demographics and the Likert-scale responses to the Usability Questionnaires (frequencies, means, and standard deviations across participants) using the Statistical Package for the Social Sciences (SPSS, v.24).

Qualitative analyses. Responses to open-ended questions on both the ES-UQs and the O-UQ were analyzed using thematic analysis (Nowell, Norris, White, & Moules, 2017), with coding informed by content analysis (guided by Schreier [2012] and Hsieh & Shannon [2005]. Author K.T.M. conducted the analysis using NVivo software (QSR International, NVivo for Mac, version 11.4.3 [2084]), coding the ES-UQs and O-UQ separately. After all open-ended responses were read, the smallest units of analysis that contained a single coherent meaning (e.g., sentence/clause, or sometimes linked sentences) were identified as separate codes. If commonalities were identified across questionnaires, the same codes were recorded. Codes were then collapsed across all six questionnaires, and amalgamated or reduced based on their correspondence with the seven ‘user experience honeycomb’ dimensions (Morville & Sullenger, 2010), plus feasibility and acceptability. Codes were compared across NDD groups. Based on review of the data by authors K.T.M., I.S., and P.C., 146 individual codes were identified and grouped into 11 categories: accessible, credible, desirable, findable, usable, useful, valuable; acceptability for NDDs; feasibility of implementation; what parents learned and experienced; and miscellaneous feedback (e.g., no comment, not applicable). The number of individual parents who endorsed each code within a category was determined, to identify whether any individual’s feedback unduly influenced a code’s predominance.

The reported results include codes endorsed by at least 5 parents for each questionnaire (one third of the final *n*).

Results

Usage

The length of time from initial log-in to the intervention to completion of Session 5 was between 4 and 14 weeks for all parents except one (ADHD), who took 26 weeks to complete. When average numbers of days to complete each session were examined for all participants (including those who did not complete all sessions), Session 2 had the longest average time to completion (10 days; however, one participant took 72 days to complete this session). Sessions 4 and 5 tended to be shortest on average (2.8 and 2.9 days respectively). Overall number of log-ins to the intervention (including log-ins to complete the session as well as to complete sleep diaries) ranged between 15 and 62 for participants who completed the entire intervention, 25 for a participant who completed 2 sessions, 35 - 42 for participants who completed 3 sessions, and 34 for one participant who only completed the first session.

Quantitative data

Parent ratings of usability, feasibility, and acceptability across all NDD diagnoses from the O-UQ and ES-UQs are presented in Table 4.5. Generally, parents' mean ratings across diagnostic groups indicated that they agreed with statements that the sessions and intervention were useful, usable/findable, accessible, credible, desirable, and valuable, with mean ratings falling between "Strongly Agree" (rating 1) and "Agree" (rating 2).

Averaged across all NDD groups, no mean ratings exceeded 3 (“Neither Agree Nor Disagree”). Ratings for the O-UQ and the ES-UQs were very similar.

In the O-UQ, average ratings across all parents showed 1) high levels of satisfaction with the intervention (between very satisfied and satisfied); 2) agreement that what was expected was present in the intervention; 3) that the intervention was feasible to implement; and 4) that the intervention was perceived as “very ready” for use by parents of children with NDDs. Eighty percent of participants said “yes” and twenty percent said “maybe” when asked about recommending the intervention to other parents of children with insomnia and the same NDD as their own children.

Parents also rated specific key features of the intervention in the ES-UQs and the O-UQ (Table 4.6), including: videos (general education videos as well as “Expert Videos,” featuring sleep experts speaking on specific topics), activities (the behavioural strategies that parents were taught and asked to implement), homework (reminders, goals, and activities for each session except Session 1), features (printable worksheets, summaries, and other supplemental materials), and support (built-in support from the intervention itself). Parents’ average ratings indicated that they agreed or strongly agreed with statements about the helpfulness of the key features. Ratings for the videos, both expert and general, tended to be more positive than for other features. Parents also generally agreed that they had adequate support, clarification, and motivation while completing the intervention, and agreement with this statement increased over sessions.

Qualitative data

Parents' responses to the open-ended questions were overwhelmingly positive (see Table 3.7) across the O-UQ and ES-UQs. Responses to the eleven categories are as follows.

Accessible: Common feedback included that sessions were easy to understand and that using the intervention was convenient and flexible to use anywhere, anytime, and with mobile devices; e.g., "I loved being able to do the program when and where I chose. I sometimes did a session all at once, other times in sections -- super convenient" (ASD parent).

Credible: A majority of parents found the intervention to be credible; for example, one ADHD parent reported, "I always felt that the professionals featured in the program were very knowledgeable on the subject and the information presented was always evidence-based and current."

Desirable: Parents perceived the visuals, graphics, and layout / design positively – for example, "Very nicely designed and looked appealing" (CP).

Findable: Overall, parents reported they could easily navigate and find information.

Usable: Parents reported that the intervention was easy to use, follow, and do at their own pace; parents provided positive feedback about the organization and presentation of the information.

Suggestions to improve the intervention's usability included: adding data input options (e.g., more drop-down menu options for bedtimes / wake times in the sleep diary), making specific changes to the website (e.g., allowing the user to stay logged in longer; having sleep diary information automatically fill some fields in later forms within the intervention), addressing technical difficulties (e.g., some parents noted that videos did not play, or that the rewards centre did not work); breaking up or shortening Session 1 (perceived to be too long).

Useful: Parents reported that session activities and content were generally useful, for example, one FASD parent reported, “It helped to better understand sleep without thinking medication is the only answer to help my children”. Some parents reported that sessions contained less useful/applicable information (particularly Sessions 1, 3 and 4), noting they were already familiar with information in Session 1.

Valuable: Parents provided general positive feedback indicating that they enjoyed participating in the intervention and the intervention had value for them. For example, parents reported comments such as, “It worked for us. My son sleeps and wakes happy and ready to take on the day. We wake and [are] not in a fog state” (CP); and “Made more progress in the weeks involved in the study than 2 years of trying to find help for my child elsewhere” (FASD).” In contrast, some parents found the intervention less valuable, reporting, “I had hoped for better success” (ADHD).

Feasibility of Implementation: Parent feedback ranged from reporting that the intervention was feasible to implement and they were able to actively use strategies, to noting that it was hard to do, apply, or find time to do. For example, one parent (ADHD) noted, “The biggest challenge we faced implementing the strategies was maintaining a consistent schedule. With shift work, after-school programs, and plans/outings with friends and family it was almost impossible to stick to a set time schedule.”

Acceptability for NDDs: To improve acceptability, parents suggested adding specific information about each NDD diagnosis and highlighting how children with

NDDs differ from TD children. For example, one parent of a child with ADHD wrote: "… in order to make this program more applicable to children with neurodevelopmental disorders I strongly feel that it needs to be modified to include background information on neurodevelopmental disorders, how they impact a child's brain (and therefore, their sleep), sleep disorders commonly seen in children with neurodevelopmental disorders and then also things to consider/additional strategies to address their sleep issues. One of the most significant things that needs to be considered is the fact that many children with neurodevelopmental disorders take medications which may further impact their sleep (e.g. stimulant medications)." However, most parents reported that information in Session 1 was generalizable to children with NDDs.

What Parents Learned and Experienced: Parents reported making progress and seeing improvements in their children's sleep, and highlighted learning the importance of consistency and routines in dealing with children's sleep problems. For example, after completing the intervention, one parent of a child with ASD reported: "I feel that parents can benefit from this regardless of their child's development. Although there were difficulties, it was in small enough steps to be very manageable."

Miscellaneous Feedback: Of note, some parents skipped most of Session 4; skipping was an option offered automatically, based on whether the child experienced the problems included in that session (night waking, napping, early morning awakenings).

Discussion and Conclusions

General Discussion

The main purpose of this study was to determine whether parents / caregivers of children with NDDs perceived the original BNBD intervention for treating insomnia in TD children to be usable, acceptable and feasible for their children with NDDs, and secondarily to determine what if any modifications were needed to adapt the intervention for children with NDDs. To do this, parents of children with NDDs (ASD, ADHD, FASD, or CP) implemented the BNBD intervention with their children, and provided feedback on its usability, acceptability, and feasibility. Overall, parents of children with NDDs found the BNBD intervention to be easy-to-use and readily applied in everyday life, and reported that it was acceptable to them as parents of children with NDDs. Even without modifications, parents reported high levels of acceptability, satisfaction, usefulness, and readiness for use with children with NDDs. Both quantitative and qualitative feedback were strongly positive in this regard, with participants providing some suggestions to make the intervention even more usable, acceptable, feasible, and appropriate.

Results showed that parents had positive user experiences, with questionnaire ratings indicating that they generally strongly agreed or agreed with statements about the intervention’s usefulness, usability, desirability (e.g., visual appeal and organization / presentation of information), accessibility (device, time, convenience, and ease of understanding), credibility (trust and reputability of the information), findability (ease of navigation), and value to them. Key features of the intervention, such as sleep diaries, session summaries, session plans, and various optional features (such as a ‘Roadblocks’ section and a ‘Reward Centre’) were also rated very positively. Parents also rated the opportunity to participate by distance as a helpful and important aspect of the intervention. The results of this study also speak to the interaction of user characteristics, environment, website characteristics, website use, support, mechanisms of change, and behaviour change, as described by Ritterband and colleagues (2009), in playing a role in the usability and feasibility of an online intervention. Importantly, an examination of usage data suggests that participants who averaged more log-ins may have been less likely to complete the intervention (for example, one participant logged in 34 times to complete only the first session – perhaps indicating some technical difficulties – in contrast to another participant who completed all 5 sessions in 15 log-ins).

The results also support a design in which core intervention components for treating insomnia in children with NDDs are transdiagnostic, with options for diagnosis-specific modules or strategies as needed. Although the original BNBD intervention for TD children was unmodified, parents of children with NDDs nevertheless reported it to be generally usable, feasible, and acceptable. This finding is consistent with other literature (Rigney et al., 2018; Harvey, 2008). Parents did suggest that inclusion of more NDD-specific information would make the intervention more acceptable, useful, and ready for use (e.g., providing information about sleep in the context of an NDD, or learning how to accommodate specific NDD symptoms in implementing intervention strategies), which seems to support the use of a modular transdiagnostic treatment for insomnia in children with NDDs. Despite the varied etiologies and presentations of children with NDDs in this study, the same strategies as used for TD children were utilized and found usable, acceptable, and feasible by their parents.

Limitations

Although this study had a small sample, typically only 8 to 10 participants are considered adequate for a usability study (Kushniruk, Patel, & Cimino, 1997). However, we could not speak to potential differences between NDD groups given the small sample and the uneven numbers by diagnostic group. In planning for an RCT, sufficient power to examine potential diagnostic group differences will be important. For example, we were not able to examine whether comorbidity of children's diagnoses influenced parents' responses about the intervention, though we hope the sample is representative of families of children with NDDs given the high rates of comorbidities across NDDs. Additionally, children's NDD diagnoses were based only on parent report, including indicating the source of the diagnosis (e.g., physician, psychologist). Finally, we included parents whose children likely had a range of different severity of NDD symptoms, which, although representative of the broader NDD population, may have impacted their experiences with the intervention.

Although feedback on usability, feasibility and acceptability was overwhelmingly positive, the sample consists of parents who successfully navigated the multi-step screening process to enroll in the study, and most completed the intervention. The study may thus have appealed to parents with greater internet literacy. The user group was also generally well-educated. The data may be most representative of parents who had positive experiences with the intervention (one participant withdrew before starting because of technical difficulties, one withdrew after completing Session 3 due to a decision to resume co-sleeping with her child, and those who did not complete the intervention took longer to complete sessions). We also note that questionnaire response options were weighted in one direction in an attempt to lessen respondent confusion, which may have increased response bias.

Conclusions

Parents of children with NDDs reported the BNBD intervention to be easy to use, manageable to implement in their everyday lives, and acceptable, despite being designed for children who did not have NDD diagnoses. The results suggest positive effects when translated from TD children to children with NDDs, and across four NDD diagnoses. However, parents did indicate that diagnosis-specific information would be helpful and increase the intervention's acceptability. Such feedback is consistent with the movement towards developing modular transdiagnostic interventions in which general strategies can be utilized by different diagnostic groups, and specific modules can be selected as needed.

The current study is the final step of an empirical foundation for the development of BNBD-NDD, a modular transdiagnostic intervention for sleep problems in children with NDDs. The results of this study have informed the development of additional material for BNBD-NDD, to be tested for efficacy in an upcoming RCT.

Table 4.1

Participant Inclusion, Exclusion, and Comorbidity Criteria

Inclusion Criteria

- Primary caregiver of child aged 4 years, 0 months to 10 years, 11 months
- Live in Canada
- Regular access to high-speed internet, email, and phone/web-camera
- Fluent in English (determined by Single Item Literacy Scale (SILS))
- Child formally diagnosed with ADHD, ASD, CP, or FASD by a physician or psychologist, as reported by parents
- Child has insomnia (determined by Behavioural Insomnia Screening Questionnaire, BIQ)
- Child attends school or preschool

Exclusion Criteria

- Participant wishes to bed-share/co-sleep with child

Child has/is:

- formally diagnosed sleep disorder other than insomnia (e.g., Sleep Apnea) or significant medical disorder that interferes with sleep
- sleep-breathing problems (determined by Pediatric Sleep Questionnaire (PSQ))
- mental health disorder that required/currently requires hospitalization / residential care (not including Emergency Room visit)
- non-ambulatory / not able to turn self over in bed
- functional impairment in adaptive skills determined by caregiver report (not dry during day, not able to feed self with utensil, cannot actively participate in dressing self, cannot follow 2-step instructions, cannot express preferences using verbal language, cannot speak in at least 4-word sentences)

Child currently taking:

- anti-epileptic and/or psychotropic medication, *excluding stimulant medication*, where dose is not stable/expected to change within 6 months
- over-the-counter/natural health medications (e.g., Benadryl) *except* melatonin to treat child's sleep problems

Comorbidity Criteria

To categorize participants/children into four primary NDD groups and minimize confounding diagnoses. FASD was considered a primary grouping criterion because diagnosis requires proven exposure to alcohol.

- ADHD group: child can have CP but not ASD or FASD
- ASD group: child can have ADHD or CP but not FASD
- CP group: no comorbid NDD diagnoses
- FASD group: child can have ADHD or ASD

Table 4.2

Demographic and Descriptive Information for Participants and Their Children

	Primary NDD Group				Total
	ADHD (*n*=4)	ASD (*n* = 9)	CP (*n* = 2)	FASD (*n* = 5)	*N* = 20
Participant Demographics					
Participants' relationship to child					
Biological Mother	3	9	2		14
Adoptive Mother	1			2	3
Adoptive Father				3	3
Participants' relationship status					
Common-law relationship	1	2			3
Legally married	3	6	2	5	16
Divorced		1			1
Spouse or Partner Lives in Home					
Yes	4	8	2	5	19
No		1			1
Participant's Current Employment Status					
Full Time	2	1		3	6
Part Time	2	4	2	2	10
Homemaker (e.g., Stay at home parent)		3			3
Other		1			1
Estimated Household Income					
Prefer not to answer				1	1
$30,000 - $39,999		1			1
$40,000 - $49,999		1			1
$50,000 - $50,999		1			1
$60,000 - $79,999		3		2	5
$80,000 - $99,999		1	1	2	4
$100,000 - $124,999			1		1
$125,000 - $149,999	1	2			3
$150,000 and over	3				3
Average Number of Other Children in Home (mean; SD)	2.75 (.96)	2.11 (.78)	1.5 (.71)	1.4 (.55)	2 (0.86)
Child Demographics					
Child Gender					
Male	3	8	2	4	17
Female	1	1	0	1	3
Child Mean Age in years (SD); Range	9.98 (.65);	8.59 (1.40);	8.00 (4.10);	9.34 (1.13);	8.99 (1.57);

		Primary NDD Group				Total
		ADHD (*n*=4)	ASD (*n* = 9)	CP (*n* = 2)	FASD (*n* = 5)	*N* = 20
		9.10 – 10.60	6.50 – 10.30	5.10 – 10.90	8.30 – 10.90	5.10 – 10.90
Child Comorbid Neurodevelopmental Disorder Diagnoses						
	None	4	6	2	1	13
	ADHD		3		4	7
	Developmental coordination disorder	1	1			2
	Mild intellectual disability				1	1
Child Physical Health Disorders						
	Epilepsy		1*			1
	Asthma		1			1
	Environmental allergies				1	1
Child Mental Health Disorder Diagnoses						
	Anxiety	1				1
	Reactive attachment disorder				1	1

Note:
* Child was being treated with anti-epileptic medication but dose was stable and not expected to change within 6 months (met inclusion criteria).

Table 4.3

Completion rates (numbers of participants) for sessions and questionnaires

Completion	All NDD (20)	ADHD (4)	ASD (9)	CP (2)	FASD (5)
Session 1	20 (100%)	4 (100%)	9 (100%)	2 (100%)	5 (100%)
Session 1 UQ	20 (100%)	4 (100%)	9 (100%)	2 (100%)	5 (100%)
Session 2	19 (95%)	4 (100%)	8 (88%)	2 (100%)	5 (100%)
Session 2 UQ	18 (90%)	4 (100%)	7 (78%)	2 (100%)	5 (100%)
Session 3	18 (90%)	4 (100%)	7 (78%)	2 (100%)	5 (100%)
Session 3 UQ	17 (85%)	4 (100%)	7 (78%)	1 (50%)	5 (100%)
Session 4	15 (75%)	4 (100%)	6 (67%)	1 (50%)	4 (80%)
Session 4 UQ	15 (75%)	4 (100%)	6 (67%)	1 (50%)	4 (80%)
Session 5	15 (75%)	4 (100%)	6 (67%)	1 (50%)	4 (80%)
Session 5 UQ	15 (75%)	4 (100%)	6 (67%)	1 (50%)	4 (80%)
Overall Intervention	15 (75%)	4 (100%)	6 (67%)	1 (50%)	4 (80%)
Overall UQ	15 (75%)	4 (100%)	6 (67%)	1 (50%)	4 (80%)

Table 4.4

Descriptive Information for Participants' Report of Children's Sleep Problems

	ADHD (*n* = 4)	ASD (*n* = 9)	CP (*n* = 2)	FASD (*n* = 5)	All (*n* = 20)
Type of Sleep Problem Endorsed (# of Participants)					
Problems falling asleep	4	9	2	5	20
Problems following bedtime routines	2	5	1	5	13
Requires adults to be present to be asleep	2	4	0	3	9
Child Sleeps In Parents' Room					
Yes	0	4	0	4	8
No	4	5	2	1	12
Child Sleeps in Parents' Bed					
Yes	n/a	2	n/a	3	5
No	n/a	2	n/a	1	3
Currently Using Melatonin					
Yes	2	1	1	3	7
No	2	8	1	2	13

Table 4.5

Participant Ratings of Usability, Acceptability and Feasibility from Usability Questionnaires (UQ): Frequencies, Means, and Standard Deviations

"Overall, this intervention / this session…"	Overall UQ (*n* = 15)	Session 1 UQ (*n* = 20)	Session 2 UQ (*n* = 18)	Session 3 UQ (*n* = 17)	Session 4 UQ (*n* = 15)	Session 5 UQ (*n* = 15)
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)
Useful						
…provided information that helped me to be better understand, and would help me treat, my child's insomnia.	1.87 (.74)	2.05 (.67)	1.94 (.42)	1.88 (.78)	2.20 (.77)	1.93 (.70)
…provided information that was useful to me as a parent of a child with a neurodevelopmental disorder who has insomnia.	2.07 (.80)	2.30 (.86)	2.11 (.76)	2.12 (.86)	2.27 (.70)	2.00 (.76)
Usable / Findable						
…was user-friendly and could be navigated with ease.	1.20 (.41)	1.45 (.60)	1.23 (.46)	1.35 (.49)	1.60 (.74)	1.40 (.51)
…took a reasonable amount of time to complete.	1.53 (.83)	1.80 (.83)	1.67 (.69)	1.41 (.51)	1.47 (.64)	1.33 (.49)
Desirable						
…was visually appealing and the organization of information on the screen was clear. The *way* the information was presented (e.g.,	1.53 (.64)	1.68 (.82)	1.44 (.51)	1.65 (.70)	1.57 (.72)	1.53 (.52)

"Overall, this intervention / this session…"	Overall UQ (*n* = 15) Mean (SD)	Session 1 UQ (*n* = 20) Mean (SD)	Session 2 UQ (*n* = 18) Mean (SD)	Session 3 UQ (*n* = 17) Mean (SD)	Session 4 UQ (*n* = 15) Mean (SD)	Session 5 UQ (*n* = 15) Mean (SD)
design, colour, font, graphics) was a positive addition to my user experience.						
Valuable						
… the information provided is valuable to me.	1.60 (.63)	1.79 (.92)	1.78 (.65)	2.06 (1.06)	2.27 (.70)	1.93 (.88)
Accessible						
…I was able to access it from the device(s) I chose to use at a time and location that was convenient to me.	1.27 (.80)	1.47 (.51)	1.50 (.62)	1.52 (.87)	1.67 (.90)	1.47 (.83)
… the information provided in the was easy for me to understand.	1.27 (.46)	1.47 (.51)	1.28 (.46)	1.29 (.47)	1.53 (.64)	1.33 (.49)
Credible						
…I believe that the information provided comes from a reputable source, and I trust the information enough to feel comfortable using it to treat my child's insomnia.	1.20 (.41)	1.53 (.70)	1. 33 (.49)	1.59 (.80)	1.40 (.63)	1.33 (.49)
Acceptable						

“Overall, this intervention / this session…”	Overall UQ (*n* = 15) Mean (SD)	Session 1 UQ (*n* = 20) Mean (SD)	Session 2 UQ (*n* = 18) Mean (SD)	Session 3 UQ (*n* = 17) Mean (SD)	Session 4 UQ (*n* = 15) Mean (SD)	Session 5 UQ (*n* = 15) Mean (SD)
…The information provided was acceptable to me, as a parent of a child with a neurodevelopmental disorder and insomnia.	1.53 (.64)	1.79 (.63)	2.00 (.84)	1.88 (.70)	2.13 (.64)	1.67 (.72)
Feasible						
… I was able to fully participate by completing activities and using or implementing the strategies with my child without any major obstacle.	2.00 (.53)	1.95 (.97)	2.11 (.76)	2.35 (1.06)	2.01 (.96)	1.67 (.72)

Note:
All items were rated on a five point Likert-scale, where participants were asked to indicate their level of agreement with the statements from: 1 = strongly agree, 2 = agree, 3 = neither agree nor disagree, 4 = disagree, 5 = strongly disagree.

Table 4.6

Participant Ratings of Specific Key Features of the Intervention from the Usability Questionnaires (UQ): Frequencies, Means, and Standard Deviations

	Overall UQ (*n* = 15) Mean (SD)	Session 1 UQ (*n* = 20) Mean (SD)	Session 2 UQ (*n* = 18) Mean (SD)	Session 3 UQ (*n* = 17) Mean (SD)	Session 4 UQ (*n* = 15) Mean (SD)	Session 5 UQ (*n* = 15) Mean (SD)
VIDEOS						
I found the videos (not including the Expert Videos) in this intervention helpful.*	1.73 (.59)	--	--	--	--	--
I found the Expert Videos in this intervention helpful.*	1.73 (.80)	--	--	--	--	--
I enjoyed the videos presented in this intervention.*	1.80 (.86)	--	--	--	--	--
The videos added educational value to the intervention. *	1.73 (.70)	--	--	--	--	--
The videos in this session helped me learn the material.**	--	1.63 (.60)	1.61 (.70)	1.53 (.62)	1.80 (.77)	1.53 (.64)
ACTIVITIES						
I found the activities within each session in the intervention helpful.*	2.00 (.76)	--	--	--	--	--
I found the activities within each session in the intervention enjoyable.*	2.20 (.68)	--	--	--	--	--
The activities within the session are helpful, and I would refer to them / use them in future**	--	1.95 (.85)	2.11 (.90	2.18 (1.07)	2.13 (.64)	1.73 (.80)

	Overall UQ (*n* = 15) Mean (SD)	Session 1 UQ (*n* = 20) Mean (SD)	Session 2 UQ (*n* = 18) Mean (SD)	Session 3 UQ (*n* = 17) Mean (SD)	Session 4 UQ (*n* = 15) Mean (SD)	Session 5 UQ (*n* = 15) Mean (SD)
HOMEWORK						
I found the homework included in each session's session plan (e.g., reminders, goals, and activities to do at home) throughout the intervention helpful and manageable.	1.93 (.80)	*No homework assigned*	1.94 (.64)	1.94 (.66)	2.13 (.64)	1.73 (.70)
FEATURES						
The features in this session (including printable worksheets, summary sheets, and any supplemental materials) are helpful, and I would refer to them in future.**	--	1.95 (.78)	2.22 (.88)	2.00 (.71)	2.33 (.82)	1.93 (.96)
SUPPORT						
I felt like I had adequate support, clarification, and motivation while completing the intervention / this session.	1.80 (.86)	2.05 (.91)	1.72 (.67)	1.76 (.66)	1.80 (.94)	1.60 (.63)

Note:

All items were rated on a five point Likert-scale, where participants were asked to indicate their level of agreement with the statements from: 1 = strongly agree, 2 = agree, 3 = neither agree nor disagree, 4 = disagree, 5 = strongly disagree. "--" indicates instances where there is no value because the feature was not asked about in that questionnaire.

* Overall Usability Questionnaire (O-UQ) only

** End of Session Usability Questionnaire (ES-UQ) only

Table 4.7

Key Codes (endorsed by >5 participants) from qualitative open-ended responses from Overall and End of Session Usability Questionnaires, grouped by category

		OVERALL UQ	SESSION 1 UQ	SESSION 2 UQ	SESSION 3 UQ	SESSION 4 UQ	SESSION 5 UQ
Category		Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]
Usability	Accessible	•Convenient & flexible [5] •Easy to understand [5]	•Easy to understand [8] •Convenient & flexible [6]	•Easy to understand [7]	•Easy to understand [6]	--	•Convenient & flexible [5] •Easy to understand [5] •Multiple & mobile device friendly [5]
	Credible	•Perceive as credible [9]	•Perceive as credible [12]	•Perceive as credible [9]	--	•Perceive as credible [6]	•Perceive as credible [8]
	Desirable	•Perceive as desirable (visual, graphic, layout) [11]	•Perceive as desirable (visual, graphic, layout) [7]	•Perceive as desirable (visual, graphic, layout) [6]	•Perceive as desirable (visual, graphic, layout) [9]	•Perceive as desirable (visual, graphic, layout) [5]	•Perceive as desirable (visual, graphic, layout) [5]
	Findable	•Findable & navigable [12]	--	--	--	--	--
	Usable	•Easy to use [6] •Could do at own pace [5]	•Easy to use [9] •Change length of session [7] •Could do at own pace [6] •Organization & presentation of information –	•Easy to use [6] •Could do at own pace [5] •Easy to follow [5]	•Easy to use [6] •Organization & presentation of information – positive feedback [6]	--	•Easy to use [8] •Easy to follow [5]

	OVERALL UQ	SESSION 1 UQ	SESSION 2 UQ	SESSION 3 UQ	SESSION 4 UQ	SESSION 5 UQ
Category	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]
		positive feedback [6] •Add more input options to sleep diaries & worksheets [5] •Address technical difficulties [5] •Suggestion to make specific changes or improvements to website [5] •Easy to follow [5]				
Useful	•Activities content useful [13] •Video/graphics useful [7] •Video testimonials would be useful [6] •Some but not all content or sessions useful [5] •Email reminders helpful [5]	•Videos/graphics useful [13] •Activities & content useful [12] •Printable features useful [9] •Already knew information in session [7] •Add more specific treatment strategies & information [6] •Generally useful/helpful [5]	•Activities & content useful [11] •Videos/graphics useful [10] •Printable features useful [6] •Already knew information in session [6] •Email reminders helpful [5]	•Activities & content – useful [13] •Generally useful/helpful [8] •Videos/graphics useful [6] •Session not useful to us [5]	•Activities – did not do since session not applicable [6] •Activities & content – useful [6]	•Activities & content – useful [9] •Generally useful/helpful [6] •Useful for future (will continue to use) [6] •Ability to review sessions – helpful [5]

	OVERALL UQ	SESSION 1 UQ	SESSION 2 UQ	SESSION 3 UQ	SESSION 4 UQ	SESSION 5 UQ
Category	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]
Valuable	•Enjoyed participating [7] •Valuable – positive feedback [5]	•Enjoyed participating [5]	•Enjoyed participating [6]	•Enjoyed participating [5]	--	•Enjoyed participating [7]
Feasibility of Implementation	•Hard to implement/do/apply [5] •Feasible to implement [5]	•Awareness that practical use of intervention is affected by external factors [5] •Hard to find time to do – other demands on time [5]	•Actively using strategies & activities [5] •Feasible to implement [5]	•Feasible to implement [6] •Actively using strategies & activities [5]	--	•Adequate amount of support – positive feedback [6]
Acceptability for NDD	•Add NDD-specific information & address what makes children with NDD different than TD [5]	•Add NDD-specific information & address what makes children with NDD different than TD [10] •Current information generalizable to NDD & comorbidities [5]	•Add NDD-specific information & address what makes children with NDD different than TD [8]	--	--	--
What Participants Learned and Experienced	•Improvements in sleep or progress [8]	--	•Learned importance of consistency & routines [6]	•Improvements in sleep or progress [8]	--	•Improvements in sleep or progress [6]

	OVERALL UQ	SESSION 1 UQ	SESSION 2 UQ	SESSION 3 UQ	SESSION 4 UQ	SESSION 5 UQ
Category	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]	Codes [# PPs endorsed by]
<u>Miscellaneous Feedback</u>	--	--	--	--	•Questions not applicable because did not use most of session [5]	--

Note:
"-- " indicates that no codes were endorsed by five or more participants. The numbers in [ ] after each code indicate the number of participants who endorsed the code. Codes in each cell are ordered by most endorsed to least endorsed.

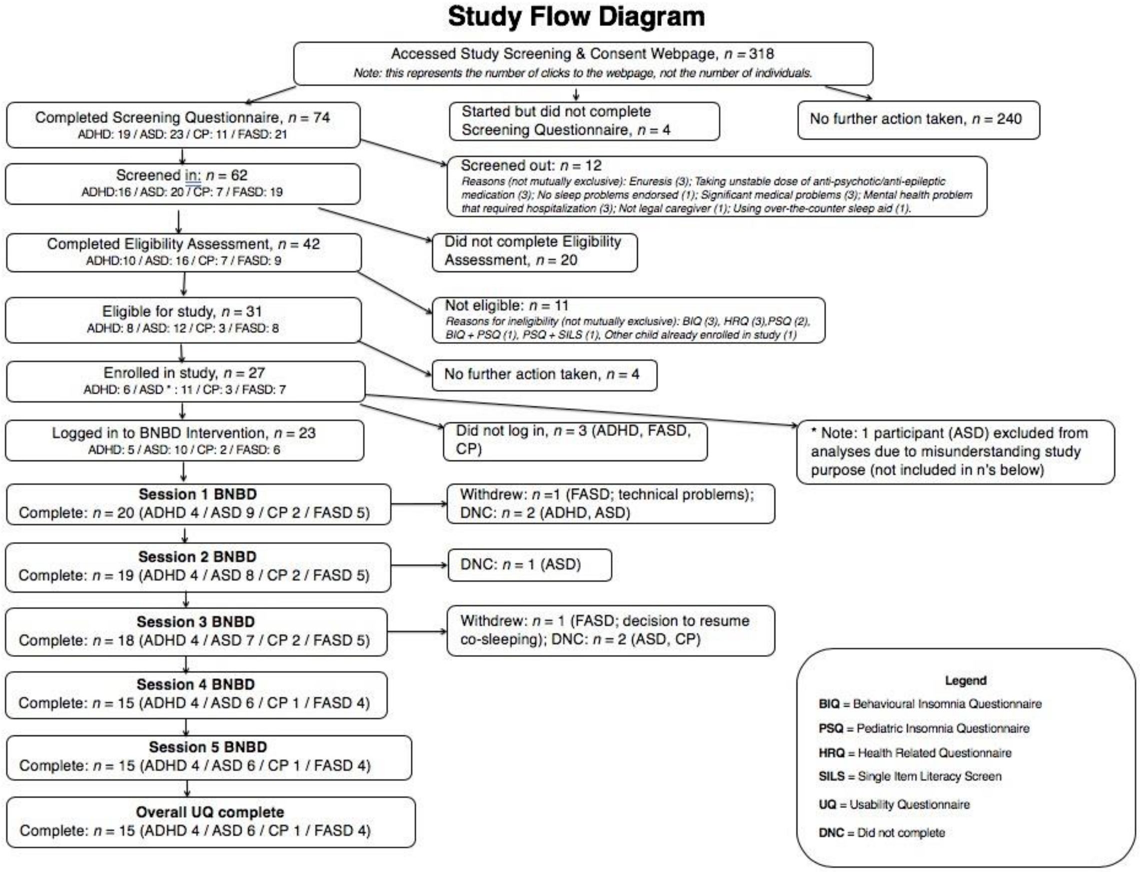

Figure 4.1. *Study Flow Diagram.*

Chapter 5: Discussion

Overview of Findings

In spite of evidence that behavioural interventions are effective for treating children's sleep problems, families experience difficulties accessing treatment, particularly families of children with NDDs. As such, the goal of the present dissertation was to explore potential means of overcoming treatment barriers and contribute to the development of an accessible sleep intervention for children with NDDs, specifically ASD, ADHD, CP, and FASD. Three studies were conducted with the following objectives: Study 1: to review the literature on online parent-implemented interventions for core NDD symptoms and associated problem behaviours in children with NDDs in order to determine whether online interventions/eHealth may be an effective way to deliver EBIs to parents of children with NDDs; Study 2: to identify and explore the barriers and facilitators to access, use, and provision of treatments for sleep problems in children with NDDs as reported by parents and front-line HCPs; and Study 3: to determine whether parents of children with NDDs perceived an eHealth parent-implemented behavioural intervention originally developed for treating insomnia in typically developing (TD) children to be usable, acceptable, and feasible.

In the following sections, I will summarize and discuss the results as they relate to each of these objectives, then review how the integrated results of these studies map onto broader theoretical and methodological concerns in the field of sleep and NDDs and online intervention research. After reviewing the strengths and limitations of the research undertaken for this dissertation, I conclude by discussing clinical implications and future directions for research.

Objective 1: To determine the state of the literature on online parent-implemented interventions for core NDD symptoms and other associated behaviours in children with NDDs, evaluate the quality of the studies examining such interventions, explore efficacy and effectiveness data, and determine whether any online parent-implemented interventions qualify as EBIs.

Although Study 1 (Chapter 2) did not focus specifically on sleep problems, it was designed to explore whether online parent-implemented interventions might be an effective way of delivering EBIs to parents of children with NDDs and thus provided a foundation to the usability study of an eHealth parent-implemented sleep intervention in Study 3. Overall, the findings of Study 1 revealed that twelve online parent-implemented interventions have been developed and tested for parents of children with ASD ($n = 8$), ADHD ($n = 3$), and FASD ($n = 1$). No interventions were found for parents of children with CP. These interventions shared some common characteristics, although both design and features were heterogenous (e.g., most interventions used a modular design and incorporated videos and slideshows; the level of external support provided to parents varied). Whereas some interventions were specifically developed for online delivery, others were adapted from evidence-based face-to-face interventions. They targeted a variety of child behavioural outcomes. Although the overall methodological quality of studies evaluating the interventions was poor, with few large-scale RCTs, under-powered analyses, high attrition, and inadequate descriptions of intervention content, it should be noted that this is an emerging field of research. Many studies were small or preliminary (e.g., pilot RCTs) and designed to pave the way for larger and higher quality RCTs. Eight of the studies demonstrated preliminary evidence of efficacy or effectiveness for improving core NDD symptoms such as social communication skills in ASD and ADHD symptoms and externalizing behaviours, as well as children's engagement in routines (ASD), parent-child interaction (ASD), and sleep (ASD). Several interventions also demonstrated improvements in parent outcomes such as stress and self-efficacy. Overall, no studies qualified as well-established EBIs, although one intervention for externalizing behaviours and ADHD symptoms in children with ADHD was considered probably efficacious (Triple P Online; Day & Sanders, 2018; Franke et al., 2017).

This systematic review contributed to the literature on online parent-implemented interventions for children with NDDs by building on and extending the findings of previous reviews. Unlike previous reviews, this review considered a range of NDD diagnoses, included only true parent-implemented interventions (rather than parent support or parent education interventions, which are designed to affect children's outcomes only indirectly), and excluded telehealth, videoconferencing, and non-online / eHealth digital interventions. The findings also had important implications for Study 3 (Chapter 4), and for the development and modification of the BNBD-NDD intervention. The majority of studies evaluating online parent-implemented interventions were methodologically poor and did not adequately describe intervention content. These findings highlight the importance of thoroughly describing both the content and means of delivery, as well as of conducting rigorous trials wherein outcomes are measured both objectively and subjectively. Furthermore, no conclusions about effectiveness can be drawn regarding the differences between interventions that were developed specifically for online delivery versus the ones that were adaptations of face-to-face interventions.

However, the review points to the importance of carefully designing and vetting online interventions through usability and feasibility testing. This is consistent with recommendations based on models that describe how online interventions can effect behaviour change (Ritterband et al., 2009). The review also showed that studies that compared self-directed with therapist-assisted intervention generally found better outcomes for children in the latter conditions. This suggests that providing external support may be an important component of online interventions, perhaps especially for parents of children with NDDs, some of whom may be stressed and find it difficult to implement strategies on their own (Hastings & Brown, 2002).

Objective 2: To identify barriers and facilitators to access, use, and provision of treatments for sleep problems in children with NDDs, as reported by their parents and the HCPs who work with them.

Results from the second study (Chapter 3) highlighted a number of key themes related to sleep treatment, and also demonstrated that the experiences of parents and HCPs were similar across different NDD diagnoses. These findings provide support for a transdiagnostic approach to sleep treatment. Findings emphasized that both parents and HCPs perceived a general lack of knowledge and awareness about sleep problems, in addition to treatments being inaccessible or limited. Both groups reported sleep problems and the treatment thereof to be especially challenging, intense, and demanding, with a high negative impact on parents’ stress, fatigue, and quality of life. As reported by participants, the perceived complexity of individualizing sleep treatment to children’s needs whilst accommodating a range of NDD symptoms and comorbidities could be overwhelming. While some parents perceived HCPs as being unhelpful or not offering enough support, HCPs reported parallel concerns about not being able to support parents adequately due to systemic constraints. They also reported worries about the feasibility of heavily burdened parents implementing sleep treatments for their children.

The study's findings emphasized that clear communication between parents and HCPs, education and awareness about sleep, fostering of positive attitudes of hopefulness and motivation, and consistent use of behavioural strategies (such as bedtime routines and healthy sleep habits) could be helpful in treating children's sleep problems. When themes from the four NDD groups were compared, few differences in barriers and facilitators emerged. The main differences related to specific concerns associated with FASD (e.g., attachment disorders / experience of trauma) and CP (e.g., pain) that could act as barriers to sleep treatment. These results support a transdiagnostic approach to treating sleep problems in children with NDDs, consistent with previous literature (e.g., Rigney et al., 2018).

To our knowledge, this is the first study to examine barriers and facilitators to accessing, providing and using sleep treatment in children with a range of NDD diagnoses. This study provides data about phenomena that have long been described anecdotally – that many parents cannot access treatment, or may not even realize that sleep problems can be treated, and that some HCPs feel that they lack the resources and knowledge to support families in treating children's sleep problems effectively. The findings of this study are also important for several other reasons. First, they can better inform our understanding of sleep treatment needs from both parents' and HCPs' perspectives. Second, they emphasize the great need for an accessible, evidence-based sleep intervention, in addition to increased awareness and advocacy for sleep problems in children with NDDs. Third, they informed the development of BNBD-NDD, a sleep intervention for children with NDDs, as described further below. As a transdiagnostic intervention, BNBD-NDD is supported by the finding that parents and HCPs utilize and recommend the same core behavioural intervention strategies across diagnoses, tailoring or customizing as needed. Finally, this study overwhelmingly highlights the fact that children's sleep problems occur in a complex psychosocial context that involves the relationship and communication between families and HCPs, and impacts parental well-being and the family as a whole.

Objective 3: To determine whether parents of children with NDDs perceive an eHealth insomnia intervention originally developed for TD children to be usable, feasible, and acceptable in its unmodified form.

The third study (Chapter 4) consisted of a usability, acceptability, and feasibility test of the BNBD intervention for insomnia in TD children, conducted with parents of children with ASD, ADHD, CP, and FASD. Results showed that parents of children with NDDs found the original, unmodified BNBD intervention to be easy to use, feasible to implement in everyday life, and acceptable even without modifications. Parents reported finding the intervention useful and ready for use with children with NDDs, and generally reported positive experiences using the intervention, even though it was developed for use with TD children. Parents provided suggestions to make the intervention even more usable, feasible, and acceptable for children with NDDs – for example, including information about sleep problems in the context of children's specific NDD diagnoses, or how to accommodate specific NDD symptoms when implementing intervention strategies.

The findings from this study have particularly important implications for developing evidence-based interventions for treating sleep in children with NDDs. Even though parents suggested that adding information about specific NDD diagnoses and modified intervention strategies would improve acceptability, they nevertheless found the intervention that was developed for TD children to be useful. Many parents reported anecdotally that they observed improvements in their children's sleep when using this intervention. The findings also lend support to the emerging evidence that pediatric sleep problems can be treated transdiagnostically (e.g., Rigney et al., 2018; Harvey, 2016; Harvey et al., 2011). Finally, the results show that parents perceived an eHealth intervention to be accessible, offering them convenience and flexibility – speaking to the promising evidence found for online parent-implemented interventions in Study 1, as well as the call for more accessible interventions emerging from the results of Study 2.

Theoretical & Methodological Implications

The cumulative findings of my dissertation have theoretical and methodological implications for the broader field of sleep and NDDs treatment research as well as the online parent-implemented intervention literature. My work focuses especially on three aspects that link the two fields. First, it addresses and explores the factors leading to families' difficulty in accessing sleep treatments, and then identifies a potential solution to overcome those barriers. Second, it addresses the need to develop an intervention for treating insomnia in children with NDDs by directly investigating families' perceived treatment needs, querying HCPs' perspectives on providing treatment, and testing an intervention and eliciting feedback from parents on the intervention's usability, feasibility, and acceptability. The usability test of the BNBD intervention with parents of children with NDDs has not only contributed to the development of BNBD-NDD, but also provides guidance for other researchers looking to modify interventions to be appropriate for children with NDDs. Third, this dissertation is highly relevant to the ongoing conversation about whether transdiagnostic interventions are a viable way of treating sleep problems in children – both TD and with NDDs, as well as across different NDD diagnoses. I will discuss the implications of this dissertation for these areas.

Barriers to Accessing Sleep Treatments: Online Parent-Implemented Interventions as a Solution

As detailed in Chapter 1, families of children with NDDs experience difficulty accessing evidence-based interventions, including early intervention and mental health services (Johnson & DeLeon, 2016). This can be related to long wait times for services and specialists, living in remote locations, and to children's NDD symptoms – for example, trying to bring a child who presents with disruptive behaviours into a clinic setting (Thomas et al., 2007). However, little research has specifically examined barriers and facilitators to sleep treatment access for families of children with NDDs.

The results of Study 2 (Chapter 3) concurred with findings by Cook, Appleton, and Wiggs (2020), who examined parent-reported barriers to seeking help and advice for sleep problems in TD children in the United Kingdom. Cook and colleagues found that parents reported several key barriers: perceiving that HCPs lacked knowledge and training about sleep, not wanting to waste either their own or HCPs' time, perceiving a lack of continuity in the information provided by HCPs (e.g., different HCPs providing conflicting information), and worrying about being judged negatively for admitting that their children had sleep difficulties. Like the parent participants in my Study 2, parents in Cook et al.'s (2020) study also found some HCPs to be unhelpful. Similarly, the findings in Study 2 lined up with a qualitative study examining the beliefs, attitudes, and perceptions of both primary caregivers and rehabilitation providers about the sleep health of children with disabilities, which found that parents lacked education on how to improve children's sleep (Chen et al., 2014). Moreover, we also know that HCPs have reported lacking time, training, and resources to provide evidence-based sleep treatments for pediatric insomnia (Boerner et al., 2014), highlighting a potential benefit of interventions that are not provided directly by HCPs.

The systematic review (Chapter 2) provided emerging evidence that online parent-implemented interventions may be an effective way to deliver EBIs to parents of children with NDDs, although more research is required. Other research has shown that parents of children with NDDs already turn to the internet to seek health information related to their children's diagnoses, yet the majority of that information consists of unsubstantiated claims or is unsupported by peer review evidence (Di Pietro, Whitely, Mizgalewicz, & Illes, 2013). An easily accessible online parent-implemented intervention may not only replace unverified online information and provide evidence-based education, but also help parents to directly improve children's sleep from home while reducing pressure for HCPs who have limited time and resources. The findings of Studies

1 and 2, added to a larger body of literature, make a compelling case for developing and exploring the efficacy of an online parent-implemented behavioural sleep intervention.

Lack of EBIs for Treating Sleep Problems in Children with NDDs: Developing BNBD-NDD

As we established in Chapter 1, despite the increasing number of studies evaluating behavioural interventions for sleep problems in children with NDDs (e.g., Hiscock et al., 2015; Papadopoulos et al., 2019), few programs are readily available to parents. Interventions for families of children with ADHD, CP, and FASD are especially scarce. At the same time, HCPs working with pediatric populations report that they lack training in sleep treatment (Bruni et al., 2004; Faruqi et al., 2011) or that they lack adequate time, resources, and funding to treat sleep problems (Honaker & Meltzer, 2016). The studies comprising this dissertation address that gap by providing evidence relevant for the development of an accessible intervention that does not require the involvement of HCPs but rather is implemented by parents.

Studies 2 and 3 informed the ongoing development of BNBD-NDD, along with information gathered by Rigney and colleagues (2018) and the expertise of HCPs elicited by Ali and colleagues (2018). From this dissertation's findings, information about treatment needs, barriers, and facilitators, as well as recommendations of parents from Study 3, were used to modify BNBD into BNBD-NDD. For example, the core intervention behavioural strategies were retained, with each of the five sessions focusing on a specific sleep problem, but the research team added information about sleep problems in the context of ASD, ADHD, CP, and FASD. In the newly developed BNBD-

NDD, parents are able to customize their pathways through the intervention by choosing to access information on their preferred NDD (and if their child has comorbid NDDs, they can access information for more than one disorder). Having access to more NDD-specific information was suggested by the participants of Study 3, and echoes the findings of Beresford et al. (2016), who found that parents of children with ASD and other NDDs who took part in a psychoeducational sleep management intervention also desired more NDD-specific information.

Among the many ways in which this dissertation contributed to the new intervention was the development of testimonial videos by parents. In Study 2, parents reported that having support – most importantly, support from other parents – was a major facilitator to their ability to access and implement sleep interventions. As such, scripts were written based on the experiences of the parent participants in Study 3, performed by actors and recorded as videos that were incorporated into BNBD-NDD. In this way, we were able to facilitate an indirect sharing of experience and support among parents (while maintaining families' anonymity). The findings of Study 3 enabled the research team to create an intervention that was responsive to parents' needs and suggestions. Having done so, the research team then moved forward with a large scale RCT evaluation of the efficacy of BNBD-NDD in children with ASD, ADHD, CP, and FASD (NCT02694003, clinicaltrials.gov). Of note, although this dissertation provides some evidence suggesting that therapist coaching or external supports may influence intervention effectiveness, the RCT will evaluate BNBD-NDD as a fully self-directed intervention. Future studies of BNBD-NDD may examine whether external support/coaching may be beneficial for some parents.

Beyond contributing to the development of BNBD-NDD, this dissertation has broader implications for the process of modifying and developing other interventions for children with NDDs. Both Studies 2 and 3 generated lists of recommended modifications to make sleep treatments more effective and appropriate for children with NDDs. Additionally, both studies demonstrated a systematic approach to the modification of an intervention for children with NDDs, beginning by establishing a balanced view of treatment needs from the dual perspectives of parents and HCPs. This was followed by a usability test of the intervention, in its original form in order to determine what needed to be changed. By eliciting feedback from parents about what would make an intervention more helpful for them, we can increase parent engagement and investment in an intervention. This modification process was theoretically informed by both the user experience honeycomb (Morville & Sullenger, 2010) and the internet intervention model of behaviour change (Ritterband et al., 2009). Contrasted with the heterogeneity and poor methodological quality of the studies identified in my systematic review (Study 1), which showed that most interventions only qualified as "possibly efficacious," the benefits of adopting a systematic and theoretically based approach to online intervention development are clear.

Can Children's Sleep Problems Be Treated Transdiagnostically?

One of the key questions woven throughout this dissertation is whether sleep problems can be treated transdiagnostically. That is, whether the same behavioural strategies that are effective with TD children can be used effectively with children with NDDs, and whether the effectiveness of these strategies generalizes across different NDD diagnoses. A body of research has suggested that insomnia is transdiagnostic, occurring in many neurodevelopmental and psychiatric disorders, and is mechanistically or biologically linked to the onset of those disorders (e.g., Dolsen, Asarnow, & Harvey, 2014; Harvey, Murray, Chandler, & Soehner, 2011). Certainly, within NDDs, sleep problems may be caused by the same biopsychosocial factors in various disorders, which are highly comorbid (Corkum et al., 2014). The presence of anxiety in children and adolescents may also be causally linked to sleep problems (Gregory & Eley, 2005; Weiner, Elkins, Pincus, & Comer, 2016), or form the third point of a triangle with NDDs and sleep problems (Hollway, Aman, & Butter, 2013; Johnson & Malow, 2008).

The results of both Studies 2 and 3 provide support for a transdiagnostic approach to treating sleep problems in children with NDDs, particularly in the themes that emerged related to implementation of treatment. In Study 2 (Chapter 3), parents and HCPs perceived a need for individualization of treatment to children's needs and NDD diagnoses, yet still used the same core behavioural strategies to treat sleep across diagnoses – a finding that appears contradictory upon first glance. However, that the actual treatment approaches and strategies used and recommended by parents and HCPs were the same across all four NDDs reflects the potential value of a transdiagnostic approach to sleep treatment, consistent with recommendations by Rigney et al. (2018) and Harvey (2009). Study 3 yielded concrete evidence of the acceptability and perceived usefulness of strategies that were developed for TD children to children with several NDD diagnoses. Based on the findings from both studies, a transdiagnostic approach would involve using core behavioural intervention strategies, but modifying and tailoring them as necessary to accommodate children's NDD symptoms and individual needs or preferences. For example, parents might develop and implement a consistent bedtime routine, but use more visual cues (e.g., visual scheduling) to support their children with NDDs. Ultimately, both studies lay a foundation for the development and potential efficacy of a transdiagnostic behavioural sleep intervention such as BNBD-NDD.

Strengths and Limitations

The studies comprising this dissertation had several strengths. Study 1 (Chapter 2) comprehensively summarized the state of the literature on online parent-implemented interventions for children with NDDs, adding novel information to a growing area of research. It also clarified some of the terminological discrepancies in the online parent-implemented intervention literature regarding telehealth versus online interventions and parent-implemented versus parent-supported interventions. Study 2 (Chapter 3) targeted and recruited a diverse sample of Canadian parents of children with NDDs and frontline HCPs who work with them. Because there were few restrictions or exclusions for parent participants, it is likely that the barriers and facilitators reported by this sample would generalize to many other parents of children with these NDDs. The sample was quite large for a qualitative study, lending further confidence to the themes that emerged from the focus groups and interviews. Study 3 (Chapter 4) utilized a combination of qualitative and quantitative methods that allowed us to collect quantitative data on the usability and feasibility of the BNBD intervention, as well as to hear directly from parents about their experiences with implementing the intervention. The combined results of Studies 2 and 3 allowed us to assess families’ treatment needs and inductively determine key barriers and facilitators. All three studies helped to bridge two areas of pediatric NDD research: sleep and online parent-implemented interventions.

Despite several strengths, there are limitations to consider. Difficulties with scheduling busy participants from different time zones in Study 2 meant that individual interviews were needed as well as focus groups, which may have affected the nature of the themes that emerged. In Study 1, the identified studies were too few and too heterogeneous to allow for a meta-analysis of efficacy / effectiveness.

Other limitations pertain to the representativeness of study samples and participants. For example, the four NDDs targeted in this dissertation are not representative of all NDDs included in the DSM-5 (American Psychiatric Association, 2013). In particular, CP stood out as different from ASD, ADHD, and FASD. Cerebral Palsy is characterized by physical disabilities and motor impairments. Consequently, sleep problems in children with CP may be more affected by factors such as pain and muscle tightness, and therefore might require additional treatment, not only behavioural intervention. This point was raised by both parent and HCP participants in Study 2, as well as by parents in Study 3. Study 2 included more participants who were parents and HCPs of children with ASD than the other three NDDs, due to the fact that the original study focused on ASD before it was expanded to include ADHD, CP, and FASD. Numbers of HCP disciplines were also not evenly distributed – perhaps a future, larger study would be more suitably powered to compare the experiences of providing sleep treatment amongst HCPs from different disciplines. Both Studies 2 and 3 (Chapters 3 and 4) relied on parent-reported formal diagnoses of NDDs, with some exclusions based on the type of HCP who diagnosed the child (according to parents). Future research would benefit from confirming NDD diagnoses. However, this would require considerable resources and would be inconsistent with the online, accessible aspect of this research. The usability study (Chapter 4) had careful inclusion and exclusion criteria regarding comorbid NDDs, to enable comparison of results from parents of children with ASD, ADHD, CP, and FASD. However, these disorders are highly comorbid (particularly ASD and ADHD, and FASD and ADHD), so future studies may want to utilize even less strict criteria about comorbidities to maximize generalizability. Finally, Studies 2 and 3 focused on recruiting parents of children with mild to moderate sleep problems and relatively high levels of adaptive functioning. Future studies could include a usability test of BNBD or BNBD-NDD for children with more severe NDD impairment (e.g., non-verbal children, or those with lower levels of cognitive or adaptive functioning) to extend the reach of the intervention.

Clinical Implications

As discussed above, the findings of this dissertation have been used to inform the development of BNBD-NDD, which, if efficacious, will become widely available to parents of children with NDDs. This research has several key clinical implications. The emergent themes from Study 2 and the feedback provided by parents in Study 3 have greatly contributed to our understanding of how behavioural interventions for sleep problems can be modified to accommodate the needs of children with NDDs. Although some of this information has previously been compiled from disparate sources in the literature (e.g., Corkum et al., 2014; Hanlon-Dearman et al., 2018; Hvolby, 2015; Jan et al., 2008; Malow et al., 2012), the current dissertation studies directly asked parents and

HCPs for their feedback and suggestions. The focus group study (Chapter 3) in particular produced specific modifications and accommodations that parents and HCPs found helpful.

These findings also highlight the importance of ensuring that HCPs understand parents' perspectives with regard to seeking and managing sleep treatments. For example, findings from Study 2 showed that many HCPs were concerned about burdening parents by asking them to implement demanding behavioural interventions. However, some parents reported finding HCPs unhelpful, unknowledgeable, and inconsistent in their recommendations (echoing the findings of Cook et al., 2020). In clinical practice, it will be very important for HCPs to consider parents' perspectives, to demonstrate empathy, support, and openness with parents around the treatment of sleep problems (Cook et al., 2020). As discussed below, ensuring that HCPs have access to accurate evidence-based sleep treatment information and training is also critical.

The current dissertation studies also make a case for the need to support parents – firstly in terms of the impact of their children's sleep problems, and secondly in the implementation of interventions. Sleep problems do not exist in isolation, but rather within a complex psychosocial context with parents bearing much of the brunt of their effects (Bernier et al., 2013; El-Sheik & Kelly, 2011; Simard-Tremblay et al., 2011). As previously discussed, parents of children with NDDs already experience higher levels of stress, lower levels of parenting self-efficacy, and poorer and less sleep themselves relative to parents of TD children (e.g., Micsinkszki, Ballantyne, Cleverley, Green, & Stremler, 2018). For example, the sleep problems of children with ASD have been shown to be associated with marital conflict and deterioration of relationships between parents, in addition to financial burden (Kirkpatrick et al., 2019). Even though online parent-implemented interventions increase accessibility and allow parents to access desired treatments to help their children, they make considerable demands of parents' time and energy. These parents are likely already tired, busy, and stressed. As discussed in Chapter 2, parental well-being and mental health can mediate children's behavioural outcomes (Sanders & Morawska, 2014). Wainer, Hepburn and McMahon Griffith (2017) suggest that when developing and evaluating parent-implemented interventions, greater attention should be paid to how the intervention can positively influence family and parent outcomes such as quality of life and self-efficacy, in order to increase parent motivation and engagement with the intervention. It would also be beneficial for future research to consider examining the wider psychosocial context of children's sleep problems, including the impact on siblings. This could be tied into cost-effectiveness research that evaluates the financial impact of children's poor sleep on the daily life of the family and on society (e.g., lost parental productivity) in relation to the costs and benefits of intervention.

Finally, this dissertation draws attention to an important aspect that cannot be overlooked: sleep problems can be difficult to treat in children with NDDs. In fact, they can be perceived by both parents and HCPs as exceptionally difficult to manage, to the extent that parents of children with ASD and ADHD believe their children's sleep problems to be more intrinsic and related to the NDD diagnosis, and less treatable, compared to beliefs held by parents of TD children (Bessey et al., 2013). How can we change inaccurate and potentially unhelpful yet pervasive beliefs? The results of Study 3 demonstrate that parents can experience success and empowerment by helping their children sleep better. Parents in the focus group study spoke of how advocating for their children's needs, and for themselves, was a powerful facilitator in the journey to seek and use treatment. This research demonstrates that researchers and clinicians should take up the torch on behalf of parents and children with NDDs. We can do so by increasing awareness of the importance of sleep, guidelines to healthy sleep, and how to access treatment for sleep. Further research on cognitive behavioural strategies to promote helpful beliefs about sleep and sleep intervention for parents may also be beneficial (Bessey et al., 2013).

Future Directions for Research

These findings could prompt additional research in numerous areas, including a greater focus on the psychosocial impact of the sleep problems of children with NDDs on their parents and families, or cost-effectiveness research (both mentioned above). Some of this research is currently taking place with the BNBD-NDD RCT. However, other clear directions for future research are well worth exploring.

Firstly, the findings of Studies 2 and 3 concur with other literature that suggest a great need for more HCP education and training in how to treat pediatric sleep problems (Boerner et al., 2014; Bruni et al., 2004; Faruqui et al., 2011; Owens, 2001). Future studies could examine whether the content of an intervention such as BNBD or BNBD-NDD could be modified for HCP use, in order to increase knowledge of how to treat sleep problems in children with NDD. In fact, several parents who participated in this dissertation research suggested in their feedback that BNBD would increase HCPs' knowledge. Potentially, HCPs who were familiar with BNBD-NDD could monitor and guide their clients in the use of BNBD-NDD should the need arise, with both parents and HCPs accessing the program simultaneously. This research would help to tackle this major barrier to treatment of sleep problems in children with NDD that was not the focus of this dissertation.

Secondly, future studies could examine how best to support intervention adherence, using BNBD-NDD or other EBIs. Adherence refers to how well an individual completes or engages with an intervention (e.g., the number of sessions completed in a modular intervention; Eysenbach, 2005). Poor adherence can mean that individuals do not receive the full "dose" of treatment. Unfortunately, self-guided online psychological treatments are plagued by low rates of adherence (Beatty & Binnion, 2016). In a systematic review of predictors and reasons for adherence to online interventions, Beatty and Binnion (2016) noted that higher treatment credibility / expectancy and guidance or support via phone or email (i.e., external support) increased adherence, whereas adherence was decreased by not having enough time, being dissatisfied with program content, perceiving content as impersonal, and having technical / computer difficulties. The finding that support can increase adherence echoes the finding of the systematic review in Chapter 2, as well as findings from the wider parent-implemented intervention literature: that therapist support could enhance outcomes in both face-to-face and online interventions (Breitenstein et al., 2014; O'Brien & Daley, 2011; Tarver et al., 2014). The question of whether support is required for an online parent-implemented intervention to be effective may be particularly salient for parents of children with NDDs, given the aforementioned elevated stress and burden. However, as noted in a review of digital parenting interventions (Breitenstein et al., 2014), coaching, feedback, and other guidance can considerably increase the cost of interventions, thus emphasizing the need for health economic analyses. A future study of BNBD-NDD could examine whether therapist guidance or external support adds value to the intervention by measuring parent satisfaction and treatment adherence. This might also allow for the exploration of whether the intervention could be extended to benefit parents of children with more severe and impairing NDD symptoms or sleep problems, perhaps with increased external supports.

Conclusion

The research presented within this dissertation is novel and contributes to the wider literature on sleep problems in children with NDDs and parent-implemented eHealth interventions. Access to EBIs is paramount, and in the present circumstances, the internet may offer a safe and effective way to bring interventions directly into families' homes. Together, these studies have contributed to the development of BNBD-NDD, an eHealth intervention for treating behavioural sleep problems in children with NDDs that is currently being tested in a Canada-wide RCT. These studies have also added to a growing body of evidence for the utility of transdiagnostic interventions for sleep problems in children with NDDs. In particular, these studies emphasize the great need to increase knowledge about healthy sleep in children with NDDs through education of both parents and HCPs. Specifically, researchers and clinicians must increase the awareness that sleep problems in children with NDDs are eminently treatable, and that healthy sleep has the potential to vastly improve children's functioning as well as parents' own quality of life. This dissertation brings together research literature, the voices and lived experiences of parents of children with NDDs and sleep problems, and the clinical experiences of HCPs who provide sleep treatment to children with NDDs. In doing so, it contributed to the development of an intervention that may help parents and their children to finally obtain many good nights' sleep.

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Appendix A. Sample Systematic Review Search Strategy

The following is a search strategy used for the PsycINFO (Ebscohost) database. Each search concept was run separately and saved. Then, all searches were combined with “AND” (i.e., ‘Concept 1 AND Concept 2 AND Concept 3 AND Concept 4’) for the final search. Concept 2 included multiple sub-concepts, which were saved and combined within it using ‘OR’ to form a final search term (e.g., ‘2a OR 2b OR 2c OR 2d OR 2e OR 2f’).

Concept 1 Children:

(DE “Pediatrics” OR TI (pediatric* OR paediatric*) OR AB (pediatric* OR paediatric*) OR SU (pediatric* OR paediatric*) OR KW (pediatric* OR paediatric*) OR MA “Child” OR TI child* OR AB child* OR SU child* OR KW child* OR TI youth* OR AB youth* OR SU youth* OR KW youth* OR MA “Adolescent” OR TI adolescen* OR AB adolescen* OR TI teen* OR AB teen* OR SU adolescen* OR SU teen* OR KW adolescen* OR KW teen* OR DE “Preschool Students” OR MA “child, preschool” OR TI preschool* OR AB preschool* OR SU preschool* OR KW preschool* OR TI toddler* OR AB toddler* OR SU toddler* OR KW toddler* OR TI minors OR AB minors OR SU minors OR KW minors OR TI kid OR AB kid OR TI kids OR AB kids OR SU kid OR SU kids OR KW kid OR KW kids OR TI (“school age child” OR “school age children”) OR AB (“school age child” OR “school age children”) OR SU (“school age child” OR “school age children”) OR KW (“school age child” OR “school age children”) OR TI (schoolchild* OR “school child” OR “school children”) OR AB (schoolchild* OR “school child” OR “school children”) OR SU (schoolchild* OR “school child” OR “school children”) OR KW (schoolchild* OR “school child” OR “school children”) OR TI juvenile* OR AB juvenile* OR SU juvenile* OR KW juvenile*)

Concept 2: Neurodevelopmental Disorders

2a. Neurodevelopmental Disorders

DE “Neurodevelopmental Disorders” OR MA “Neurodevelopmental Disorders” OR TI (“neurodevelopmental disorder” OR “neurodevelopmental disorders” OR “neurodevelopment disorder” OR “neurodevelopment disorders”) OR AB (“neurodevelopmental disorder” OR “neurodevelopmental disorders” OR “neurodevelopment disorder” OR “neurodevelopment disorders”) OR SU (“neurodevelopmental disorder” OR “neurodevelopmental disorders” OR “neurodevelopment disorder” OR “neurodevelopment disorders”) OR KW (“neurodevelopmental disorder” OR “neurodevelopmental disorders” OR “neurodevelopment disorder” OR “neurodevelopment disorders”) OR TI “neurodevelopment* disorder*” OR AB “neurodevelopment* disorder*” OR SU “neurodevelopment* disorder*” OR KW “neurodevelopment* disorder*” OR TI (“neurodevelopmental disability” OR “neurodevelopmental disabilities” OR “neurodevelopment disability” OR “neurodevelopmental disabilities”) OR AB (“neurodevelopmental disability” OR “neurodevelopmental disabilities” OR “neurodevelopment disability” OR “neurodevelopmental disabilities”) OR SU (“neurodevelopmental disability” OR “neurodevelopmental disabilities” OR “neurodevelopment disability” OR “neurodevelopmental disabilities”) OR KW (“neurodevelopmental disability” OR “neurodevelopmental disabilities” OR “neurodevelopment disability” OR “neurodevelopmental disabilities”) OR TI NDD OR AB NDD OR SU NDD OR KW NDD OR TI “neuropsychological disorder*” OR AB “neuropsychological disorder*” OR SU “neuropsychological disorder*” OR KW “neuropsychological disorder*” OR TI (“neuropsychological disability” OR “neuropsychological disabilities”) OR AB (“neuropsychological disability” OR “neuropsychological disabilities”) OR SU (“neuropsychological disability” OR “neuropsychological disabilities”) OR KW (“neuropsychological disability” OR “neuropsychological disabilities”) OR TI (“developmental disorder*” OR “development disorder*”) OR AB (“developmental disorder*” OR “development disorder*”) OR SU (“developmental disorder*” OR “development disorder*”) OR KW (“developmental disorder*” OR “development disorder*”) OR DE “Developmental Disabilities” OR MA “Developmental Disabilities” OR TI (“developmental disabilit*” OR “development disabilit*”) OR AB (“developmental disabilit*” OR “development disabilit*”) OR SU (“developmental disabilit*” OR “development disabilit*”) OR KW (“developmental disabilit*” OR “development disabilit*”) OR TI (“developmental delay” OR “developmentally delayed”) OR AB (“developmental delay” OR “developmentally delayed”) OR SU (“developmental delay” OR “developmentally delayed”) OR KW (“developmental delay” OR “developmentally delayed”))

2b. ADHD

(DE “Attention Deficit Disorder with Hyperactivity” OR DE “Attention Deficit Disorder” OR MA “Attention Deficit Disorder with Hyperactivity” OR TI (“attention deficit disorder with hyperactivity” OR “attention deficit hyperactivity disorder” OR “attention deficit disorder” OR “attention deficit” OR hyperkinesis”) OR AB (“attention deficit disorder with hyperactivity” OR “attention deficit hyperactivity disorder” OR “attention deficit disorder” OR “attention deficit” OR hyperkinesis”) OR SU (“attention deficit disorder with hyperactivity” OR “attention deficit hyperactivity disorder” OR “attention deficit disorder” OR “attention deficit” OR hyperkinesis”) OR KW (“attention deficit disorder with hyperactivity” OR “attention deficit hyperactivity disorder” OR “attention deficit disorder” OR “attention deficit” OR hyperkinesis”) OR TI ADHD OR AB ADHD OR SU ADHD OR KW ADHD)

2c. ASD

(TI autism OR AB autism OR SU autism OR KW autism OR DE “Autism Spectrum Disorders” OR TI (“Autism Spectrum Disorder*” OR “Autism Spectrum”) OR AB (“Autism Spectrum Disorder*” OR “Autism Spectrum”) OR SU (“Autism Spectrum Disorder*” OR “Autism Spectrum’) OR KW (“Autism Spectrum Disorder*” OR “Autism Spectrum”) OR MA “autistic disorder” OR TI (“autistic disorder*” OR autistic) OR AB (“autistic disorder*” OR autistic) OR SU (“autistic disorder*” OR autistic) OR KW (“autistic disorder*” OR autistic) OR TI ASD OR AB ASD OR SU ASD OR KW

ASD OR TI Asperger* OR AB Asperger* OR SU Asperger* OR KW Asperger* OR MA "Asperger syndrome" OR TI ("Asperger syndrome" OR "Asperger's syndrome" OR AB ("Asperger syndrome" OR "Asperger's syndrome") OR SU ("Asperger syndrome" OR "Asperger's syndrome") OR KW ("Asperger syndrome" OR "Asperger's syndrome") OR MA "child development disorders, pervasive" OR TI ("pervasive developmental disorder*" OR "pervasive development disorder*" OR "pervasive development*") OR AB ("pervasive developmental disorder*" OR "pervasive development disorder*" OR "pervasive development*") OR SU ("pervasive developmental disorder*" OR "pervasive development disorder*" OR "pervasive development*") OR KW ("pervasive developmental disorder*" OR "pervasive development disorder*" OR "pervasive development*") OR TI PDD OR AB PDD OR SU PDD OR KW PDD)

2d. CP

(DE "Cerebral Palsy" OR MA "Cerebral Palsy" OR TI "cerebral palsy" OR AB "cerebral palsy" OR SU "Cerebral Palsy" OR KW "Cerebral Palsy" OR TI "CP" OR AB "CP" OR SU "CP" OR KW "CP")

2e. FASD

(MA "fetal alcohol spectrum disorders" OR TI ("fetal alcohol spectrum disorder*" OR "foetal alcohol spectrum disorder*" OR "fetal alcohol" OR "foetal alcohol") OR AB ("fetal alcohol spectrum disorder*" OR "foetal alcohol spectrum disorder*" OR "fetal alcohol" OR "foetal alcohol") OR SU ("fetal alcohol spectrum disorder*" OR "foetal alcohol spectrum disorder*" OR "fetal alcohol" OR "foetal alcohol") OR KW ("fetal alcohol spectrum disorder*" OR "foetal alcohol spectrum disorder*" OR "fetal alcohol" OR "foetal alcohol") OR TI FASD OR AB FASD OR SU FASD OR KW FASD OR TI ("Fetal alcohol spectrum" OR "foetal alcohol spectrum") OR AB ("Fetal alcohol spectrum" OR "foetal alcohol spectrum") OR SU ("fetal alcohol spectrum" OR "foetal alcohol spectrum") OR KW ("fetal alcohol spectrum" OR "foetal alcohol spectrum") OR DE "Fetal Alcohol Syndrome" OR TI ("fetal alcohol syndrome" OR "foetal alcohol syndrome") OR AB ("fetal alcohol syndrome" OR "foetal alcohol syndrome") OR SU ("fetal alcohol syndrome" OR "foetal alcohol syndrome") OR KW ("fetal alcohol syndrome" OR "foetal alcohol syndrome") OR TI "FAS" OR AB "FAS" OR SU "FAS" OR KW "FAS" OR TI ("alcohol related neurodevelopmental disorder*" OR "alcohol related neurodevelopment disorder" OR "alcohol related neurodevelopmental" OR "alcohol related neurodevelopment") OR AB ("alcohol related neurodevelopmental disorder*" OR "alcohol related neurodevelopment disorder" OR "alcohol related neurodevelopmental" OR "alcohol related neurodevelopment") OR SU ("alcohol related neurodevelopmental disorder*" OR "alcohol related neurodevelopment disorder" OR "alcohol related neurodevelopmental" OR "alcohol related neurodevelopment") OR KW ("alcohol related neurodevelopmental disorder*" OR "alcohol related neurodevelopment disorder" OR "alcohol related neurodevelopmental" OR "alcohol related neurodevelopment") OR TI ("fetal alcohol effects" OR "foetal alcohol effects") OR AB ("fetal alcohol effects" OR "foetal alcohol effects") OR SU ("fetal alcohol effects" OR "foetal alcohol effects") OR KW ("fetal alcohol effects" OR "foetal alcohol effects") OR

TI “FAE” OR AB “FAE” OR SU “FAE” OR KW “FAE” OR TI “prenatal alcohol exposure” OR AB “prenatal alcohol exposure” OR SU “prenatal alcohol exposure” OR KW “prenatal alcohol exposure” OR TI “PAE” OR AB “PAE” OR SU “PAE” OR KW “PAE” OR TI (“fetal alcohol exposure” OR “foetal alcohol exposure”) OR AB (“fetal alcohol exposure” OR “foetal alcohol exposure”) OR SU (“fetal alcohol exposure” OR “foetal alcohol exposure”) OR KW (“fetal alcohol exposure” OR “foetal alcohol exposure”))

2f. Other neurodevelopmental disorders

(DE “Communication Disorders” OR MA “Communication Disorders” OR TI (“Communication disorder*” OR “communicative disorder*”) OR AB (“Communication disorder*” OR “communicative disorder*”) OR SU (“Communication disorder*” OR “communicative disorder*”) OR KW (“Communication disorder*” OR “communicative disorder*”) OR MA “social communication disorder” OR TI (“social communication disorder*” OR “social communicative disorder*”) OR AB (“social communication disorder*” OR “social communicative disorder*”) OR SU (“social communication disorder*” OR “social communicative disorder*”) OR KW (“social communication disorder*” OR “social communicative disorder*”) OR TI “intellectual disorder*” OR AB “intellectual disorder*” OR SU “intellectual disorder*” OR KW “intellectual disorder” OR MA “Intellectual Disability” OR TI (“intellectual 188isability*” OR “intellectually disabled”) OR AB (“intellectual 188isability*” OR “intellectually disabled”) OR SU (“intellectual 188isability*” OR “intellectually disabled”) OR KW (“intellectual 188isability*” OR “intellectually disabled”) OR DE “Intellectual Development Disorder” OR TI “intellectual development disorder*” OR AB “intellectual development disorder*” OR SU “intellectual development disorder*” OR KW “intellectual development disorder*” OR DE “Delayed Development” OR TI (“developmental delay” OR “developmentally delayed”) OR AB (“developmental delay” OR “developmentally delayed”) OR SU (“developmental delay” OR “developmentally delayed”) OR KW (“developmental delay” OR “developmentally delayed”))

Concept 3: Intervention

DE “Intervention” OR TI intervention* OR AB intervention* OR SU intervention* OR KW intervention* OR “DE “Treatment” OR TI treatment* OR AB treatment* OR SU treatment* OR KW treatment* OR TI therap* OR AB therap* OR SU therap* OR KW therap* OR TI (program OR programs) OR AB (program OR programs) OR SU (program OR programs) OR KW (program OR programs))

Concept 4: Online / eHealth

(TI online OR AB online OR DE “Online Therapy” OR SU online OR KW online OR TI web OR AB web OR DE “Websites” OR SU online OR KW web OR TI website OR AB website OR SU website OR KW website OR DE “Internet” OR TI internet OR AB internet OR MA “Internet” OR SU internet OR KW internet OR DE “Telemedicine” OR MA “Telemedicine” OR TI (“e-health” OR “ehealth” OR telehealth OR mhealth OR “mhealth” OR “mobile health”) OR AB (“e-health” OR “ehealth” OR telehealth OR mhealth OR “m-health” OR “mobile health”) OR SU (“e-health” OR “ehealth” OR telehealth OR mhealth OR “mobile health”) OR KW (“e-health” OR “ehealth” OR telehealth OR mhealth OR “m-health” OR “mobile health”) OR TI virtual OR AB virtual OR SU virtual OR KW virtual OR DE “Electronic Learning” OR TI (“e-learning” OR elearning) OR AB (“e-learning” OR elearning) OR SU (“e-learning” OR elearning) OR KW (“e-learning” OR elearning) OR TI (“web-based” OR “web based”) OR AB (“web-based” OR “web based”) OR SU (“web-based” OR “web based”) OR KW (“web-based” OR “web based”) OR TI (“internet-based” OR “internet based”) OR AB (“internet-based” OR “internet based”) OR SU (“internet-based” OR “internet based”) OR KW (“internet-based” OR “internet based”))

Appendix B. Study 2 Brief Eligibility Screening Questionnaire for Parents

Item #	*Question*	*Response options*
1	Are you the parent of a child between the ages of 4 and 12 years with a diagnosis of one of the following neurodevelopmental disorders (please select which one(s): a) Autism Spectrum Disorder (ASD) b) Attention-Deficit/Hyperactivity Disorder (ADHD) c) Fetal Alcohol Spectrum Disorder (FASD) d) Cerebral Palsy (CP) e) No	[Select one of the options]
2	Has your child ever had behavioural sleep problems? Please select all that apply. a) Problems following bedtime routines b) Difficulty obeying parents' requests c) Resistance to parents' requests d) Difficulty following bedtime schedule e) Needing constant reminders for each task during bedtime routine f) Problems falling asleep g) Lying awake in bed after lights out for more than 20 minutes h) Calling out that he/she can't fall asleep i) Getting out of bed j) Crying at bedtime k) Problems staying asleep l) Often waking up during the night and difficulty falling back asleep m) Waking up parents to help him/her fall asleep n) Sneaking into parents' bed o) No – my child does not have / has not had behavioural sleep problems.	[Checkbox]
3	Has your child ever been diagnosed with a sleep disorder by a health professional? *If "Yes," branch:* Please select from the following drop-down menu: a) Insomnia b) Sleep Apnea c) Restless Leg Syndrome d) Periodic Limb Movement Disorder e) Sleepwalking f) Sleep Terrors g) Sleep Talking h) Narcolepsy i) Other sleep disorder {Textbox for written description}	Yes/No *Branch:* a) through i)
4	Have you and your child lived in Canada for at least six months?	Yes/No
5	How would you describe the community where you live? a) Rural (population less than 10,000) – close to an urban centre	a) / b) / c) / d) / e)

	b) Rural (population less than 10,000) – in a remote location c) Town d) City (population less than 500,000) e) City (population more than 500,000)	
6	Can you answer written questions in English and take part in an online discussion in English?	Yes/No
7	What province/territory do you live in?	Dropdown menu: Alberta, British Columbia, Manitoba, New Brunswick, Newfoundland & Labrador, Northwest Territories, Nova Scotia, Nunavut, Ontario, Prince Edward Island, Quebec, Saskatchewan, Yukon Territory.
8	Do you have access to a computer (desktop or laptop) with: - secure, private Internet access (e.g., not public wifi), - a web camera, - and a microphone? *If Yes, Branch to:* Please select whether your Internet connection is grounded (Ethernet), wireless, or both: • Grounded (Ethernet) • Wireless • Both	Yes/No *Branch if Yes:* Checkbox
9	How did you hear about this study?	{List with checkboxes: Google search, Referral from health professional [insert drop down list of the type of health professional – family doctor, pediatrician, psychologist, other (textbox)], word-of-mouth [insert drop down list of family member, friend, colleague, other (textbox)], national advertisement [insert textbox for name], local advertisement [insert textbox for name], my child's school, my child's daycare, Facebook, Twitter, magazine [insert text box for name of magazine], Other (insert text box)}
10	Please provide your contact information. We will email you to let you know if you are eligible for the study. Preferred method of contact? • Email • Phone	{Checkboxes} {Textboxes}

	a. First name: _________ b. Email address: _______________ c. Phone (optional): __________	

Click here to **<u>Submit responses</u>**

<u>Scoring Criteria for eligibility</u>
Participants' responses must match the following:
1 – Yes – one of the disorders selected
2 – any of a) to n) are checked off. [Not eligible if o) is checked off.]
3 – Not eligible if selects Yes + any of b) through i). a) (Insomnia) is acceptable.
4 – any of a), b), c), d), or e). We may use this as a screening criteria to specifically target people from rural populations as recruitment commences.
5 – Yes.
6 - any response from drop-down menu. This will be used as a grouping criteria for placing participants into the correct regional focus group.
7- Yes. [Not eligible if No.] Grounded (Ethernet-based) internet connections will be preferred for focus group participants (faster for Blackboard Collaborate).

Appendix C. Study 2 Full Eligibility Screening Questionnaire for Parents

General Information

1. Please provide your contact information. We will primarily use email to contact you.
   a. First name: _________
   b. Email address: ________
   c. Phone number (optional): ________
2. How did you hear about this study? Check all that apply.
   a. {List with checkboxes: Google search, Referral from health professional [insert drop down list of the type of health professional – family doctor, pediatrician, psychologist, other (textbox)], word-of-mouth [insert drop down list of family member, friend, colleague, other (textbox)], national advertisement [insert textbox for name], local advertisement [insert textbox for name], my child's school, my child's daycare, Facebook, Twitter, magazine [insert text box for name of magazine], Other (insert text box)}
3. What is your postal code?
4. What is your age? {Drop down menu of numbers}
5. What is your child's birth date? (mm/yy)
6. Does your child have one of the following diagnosed neurodevelopmental disorders:
   a. Autism Spectrum Disorder (ASD) {Yes/No}
   b. Attention-Deficit/Hyperactivity Disorder (ADHD) {Yes/No}
   c. Fetal Alcohol Spectrum Disorder (FASD) {Yes/No}
   d. Cerebral Palsy (CP) {Yes/No}
7. If you selected more than one of the above neurodevelopmental disorders (i.e., your child has multiple diagnoses), please select which disorder you would prefer to discuss in the focus group:
   a. ASD
   b. ADHD
   c. FASD
   d. CP

If your child has multiple diagnoses, please answer the following questions about the primary diagnosis of preference you selected in question 7.

8. When was the diagnosis made?{Dropdown menu showing years}
9. Who diagnosed your child? {Dropdown menu}
   a. Physician
      i. Family doctor

        ii. Paediatrician
        iii. Pediatric sub-specialist
        iv. Neurologist
    b. Clinical psychologist
    c. Psychiatrist
    d. Other {open text-box}

In order to figure out whether you are eligible to participate in the focus groups, we need to learn a little bit more about your child's behavioural insomnia. The following questions ask about your child's bedtime and sleep. When answering these questions, please consider what your child's behaviour is usually like in a **typical week** over the **past month**. If your child's behaviour has been unusual for a specific reason (e.g., sickness, away on vacation) during this time, choose the most appropriate answer that captures your child's **typical** behaviour.

Behavioural Insomnia Screening Questionnaire

Cluster A: Bedtime Resistance/Sleep Onset

1. A) After saying goodnight, on average, how many times a night (before your child falls asleep) do you have to go back to your child as a result of a protest from your child (e.g., crying, bids to stay up longer, avoidance strategies, fussing or becoming upset). We call this a reunion (i.e., when you have to go to your child to respond to, or manage, their protest).
    a. 0 reunions
    b. 1 reunion
    c. 2 reunions
    d. 3 or more reunions

   B) How many nights per week do these protests happen?
    a) 0 nights/week
    b) 1 night/week
    c) 2 nights/week
    d) More than 2 nights/week
    e) Every night

2. How long does your child take to fall asleep once in bed?
    a. 1-10 minutes
    b. 11-19 minutes
    c. 20 minutes or longer

3. How many nights per week do you remain in your child's room until he/she falls asleep? (This could include rocking, lying with, or rubbing your child's back to get your child to fall asleep at bedtime)
    a. 0 nights/week

    b. 1 night/week
    c. 2 nights/week
    d. More than 2 nights/week
    e. Every night

4. For how long has your child had problems settling to sleep?
    a. Less than 1 month
    b. 1 month
    c. More than 1 month

<u>Cluster B: Night Waking</u>

1. AFTER your child has fallen asleep and has been sleeping for at least 10 minutes, how many times per night does he/she awake and need your help to fall back to sleep (e.g., lie down with him/her; allow him/her to sleep in your bed; rub your child's back or something similar)?
    a. 0
    b. 1
    c. 2
    d. 3 or more times

2. AFTER your child has fallen asleep and has been asleep for at least 10 minutes, and wakes, how long does your child's individual night awakenings usually last?
    a. Less than 10 minutes
    b. 10-19 minutes
    c. 20 minutes or longer

3. AFTER your child has fallen asleep and has been asleep for at least 10 minutes, how many nights <u>a week</u> does your child awaken and need your help to fall back to sleep?
    a. 0 nights/week
    b. 1 night/week
    c. 2 nights/week
    d. 3 nights/week or more
    e. every night

4. For how long have your child's night wakings been occurring?
    a. Less than 1 month
    b. 1 month
    c. More than 1 month

<u>Additional Questions</u>

1) Do you believe your child wakes up too early in the morning?
    - Yes
    - No

2) On the provided scale rate your opinion of the following:

		0 = Low	1 = Low/ Medium	2 = Medium	3 = Medium/ High	4 = High	N.A.
a.	**The severity of your child's sleep problem**						▨
b.	**The negative impact of your child's sleep problem on him/her in terms of:**						▨
	i. Fatigue/malaise (a general feeling of discomfort or uneasiness whose can't be easily identified)						▨
	ii. Attention, concentration, or memory impairment						▨
	iii. Impaired social, family, vocational, or academic performance						
	iv. Mood disturbance/ irritability						▨
	v. Daytime sleepiness						▨
	vi. Behavioural problems (e.g., hyperactivity, impulsivity, aggression)						▨
	vii. Reduced motivation/energy/initiative						▨
	viii. Proneness for errors/accidents						▨
	ix. Concerns or worries about sleep						▨
c.	**The negative impact of your child's sleep problem on the rest of the family**						▨

d) Please indicate whether you believe the following statement to be true or false in regard to your child's sleep problem:

> His/Her sleep problem occurs despite adequate opportunity (i.e., enough time is allotted for sleep) and circumstances (i.e., the environment is safe, dark, quiet, and comfortable) for sleep. {TRUE/FALSE}

3) Does your child sleep in your room?

☐ Yes
☐ No

4) Does your child sleep in your bed?

☐ Yes

☐ No

5) Does your child have a television in his or her bedroom?
   ☐ Yes
   ☐ No

6) Does your child have a computer in his or her bedroom?
   ☐ Yes
   ☐ No

7) If your child is between the ages of 6-12, how late do they stay up in the evening watching television or playing video games?

   {Text box to insert # hours}

☐ Child is not between 6-12 years of age

Pediatric Sleep Questionnaire

(rule out presence of sleep apnea)

1. Does your child snore during more than 3 sleeps per week (this includes night sleeps and naps)? {Yes/No}

If "Yes," branch to following questions:

2. While sleeping, does your child ever snore? {Yes/No}
3. While sleeping, does your child snore more than half the time? {Yes/No}
4. While sleeping, does your child always snore? {Yes/No}
5. While sleeping, does your child snore loudly? {Yes/No}
6. While sleeping, does your child have "heavy" or loud breathing?"{Yes/No}
7. While sleeping, does your child have trouble breathing, or struggle to breathe? {Yes/No}

[Scoring: Yes = 1, No = 0. Average score over 6 items (i.e., sum the number of 1s and divide by 6). Scores > 0.33 indicates abnormality.]

SCORING/ELIGIBILITY CRITERIA

General Information Section

- ensure that child is between 1-12 years (based on provided age (month/year))
- ensure that child was diagnosed by one of the listed health professionals (e.g., not by a "naturopath" or a non-formal diagnosis, something that might be listed by parent in the "Other" textbox) – if parent lists an unacceptable 'Other' profession, they will be excluded from the study
- ensure that postal code matches with province of residence provided by parent in previous brief eligibility questionnaire

Behavioural Insomnia Screening Questionnaire – Scoring Criteria

Because we are recruiting parents of children with a formal diagnosis of one of the three NDD groups, we assume that all children will be greater than 24 months in age (up to age 12). Based on this, we will use the scoring criteria for >24 months age as a minimum.

<u>Cluster A: Bedtime Resistance/Sleep Onset – Scoring Criteria:</u>

Child is considered having a sleep disturbance in this area (i.e., <u>**Bedtime Resistance/Sleep Onset)**</u> if the child meets <u>**two of the three**</u> of the following criteria:

1) More than three reunions for 12-24 month olds/more than two reunions for >24 month olds occur <u>two or more nights per week</u> (i.e., bids, protests, struggles) (Q1A & B)
2) >30 minutes to fall asleep for 12-24 month olds/> 20 minutes to fall asleep for >24 month olds (Q2)
3) Parent remains in room for sleep onset for two or more nights per week (Q3) <u>AND</u>
4) The episodes have been occurring for one or more months (Q4)

(NB - based on Anders & Dahl 2007 Table 18-3 for Sleep onset dyssomnia - Disturbance)

	Anders & Dahl age category	Age 12-24 months	Age >24 months	Age >36 months
Reunions		More than 3 reunions (resistances going to bed (e.g., repeated bids, protests, struggles) at least 2 nights/week	More than 2 reunions (resistances going to bed (e.g., repeated bids, protests, struggles) at least 2 nights/week.	More than 2 reunions (resistances going to bed (e.g., repeated bids, protests, struggles)) at least 2 nights/week
	?aire item # & (response)	1 A – (e) or above AND 1B – (b) or above	1A – (d) or above AND 1B – (b) or above	1A – (b) or above AND 1B – (b) or above
Duration of sleep onset		> 30 min	> 20 min	> 20 min
	?aire item # & (response)	2 (d or above)	2 (c or above)	2 (c or above)

Parental Presence		Remains in room at least 2 nights/week	Remains in room at least 2 nights/week	Remains in room at least two nights/week
	?aire item # & (response)	3 (c or above)	3 (c or above)	3 (c or above)
SUMMARY		2 or 3 of above + Duration of problem	2 or 3 of above + Duration of problem	2 or 3 of above + Duration of problem
# of episodes / week		2 or more	2 or more	2 or more
Duration of problem		1 month or more	1 month or more	1 month or more
	?airre item # & (response)	4 (b or above)	4 (b or above)	4 (b or above)

Cluster B: Night Waking – Scoring Criteria:

Child is considered having a sleep disturbance in this area (i.e., **Night Waking)** if the child has **both** of the following:

1) 3 or more awakenings per night for 12-24 month olds/2 or more awakenings a night for >24-35month olds, 2 or more awakenings a night for >36 month olds (Q1)
2) the awakenings (in total) last for 30 minutes or more for 12-24 month olds/20 minutes or more for >24-35 month olds/10 minutes or more for >36 month olds (Q2 x Q1)
AND
3) The episodes occur **two or more times** per week (Q3) for **one month** or more (Q4)

NB - based on Anders & Dahl 2007 Table 18-2 for Night waking dyssomnia - Disturbance

		BNBD Trial	BNBD Trial	BNBD Trial
	Anders & Dahl age category	Age 12-24 months	Age >24 months	Age >36 months
# of wakings requiring parental presence		3 or more	2 or more	2 or more
	?aire item # & (response)	1 (d or above)	1 (c or above)	1 (c or above)

Duration of Wakings (total)		>/= 30 min (total)	>/= 20 min (total)	>/= 10 min (total)
	?aire item #	IF (#1 * #2) >/= 30 min	IF (#1 * #2) >/= 20 min	IF (#1 * #2) >/= 10 min
# of episodes / week		2 or more per week	2 or more per week	2 or more per week
	?aire item # & (response)	3 (b or above)	3 (b or above)	3 (b or above)
SUMMARY		Must meet all 3 + Duration of problem	Must meet all 3 + Duration of problem	Must meet all 3 + Duration of problem
Duration of problem		1 month or more	1 month or more	1 month or more
	?aire item # & (response)	4 (b or above)	4 (b or above)	4 (b or above)

Additional Questions

Used to assess criteria for ICSD-3/DSM-5 insomnia; score based on frequency of responses.

Appendix D. Study 2 Eligibility Screening Questionnaire for HCPs

Item #	*Question*	*Response options*
1	Please select your profession: • Physician > Family physician / general practitioner > Paediatrician > Paediatric subspecialist > Neurologist > Psychiatrist • Nurse • Clinical Psychologist • Social Worker • Occupational Therapist If you selected "paediatric subspecialist," please specify: [text-box]	{Drop-down menu}
2	Are you a credentialed independent practitioner?	Yes/No
3	Have you been practicing in Canada for at least six months? *If Yes, branch to:* How long have you been practicing in Canada? ___ Months / ___ Years	Yes/No {Drop-down menu for each of months (1-12)/years(1-50)}
4	In which province/territory do you practice? • Alberta • British Columbia • Manitoba • New Brunswick • Newfoundland & Labrador • Northwest Territories • Nova Scotia • Nunavut • Ontario • Prince Edward Island • Quebec • Saskatchewan • Yukon Territory	{Drop-down menu}
5a	As part of your practice, do you see children ages 4 - 12 years-old who have been diagnosed with any of the following neurodevelopmental disorders? Please select all of the ones you see.	[Check box] a) Autism Spectrum Disorder (ASD) b) Attention-Deficit / Hyperactivity Disorder (ADHD)

		c) Fetal Alcohol Spectrum Disorder (FASD) d) Cerebral Palsy (CP) e) No
5b	*If Yes to 5a, branch to:* Are any of the neurodevelopmental disorders that you selected areas in which you specialize? If yes, which ones?	Yes/No Checkbox for ASD, ADHD, FASD, CP
5c	*If Yes to 5a, branch to:* Do any of your 4 – 12 year-old patients with neurodevelopmental disorders experience behavioural sleep problems (such as bedtime resistance, difficulty falling/staying asleep, frequent awakenings) or have their families sought treatment/advice for behavioural sleep problems? If yes, which populations do you see these sleep problems in?	Yes/No *If yes, branch to:* Checkbox: ASD ADHD FASD CP
6	If you were to participate in a focus group discussion on sleep problems in one of the neurodevelopmental disorder populations you indicated that you work with, which one would you prefer to discuss (e.g., ASD, ADHD, FASD, or CP)? Please select **only one group**.	a) ASD b) ADHD c) FASD d) CP *If possible – have branching so it leads only from previous options selected*
7	Are you able to complete a series of English-language questionnaires and participate in an online discussion in English?	Yes/No
8	Do you have access to a computer (desktop or laptop) with: - secure, private Internet accses (e.g., not public wifi), - a web camera, - and a microphone? *If Yes, Branch to:* Please select whether your Internet connection is grounded (Ethernet), wireless, or both: • Grounded (Ethernet) • Wireless • Both	Yes/No *Branch if Yes:* Checkbox
9	How did you hear about this study?	{List with checkboxes: Google search, From a

		client/patient, word-of-mouth [insert drop down list of family member, friend, colleague, other (textbox)], national advertisement [insert textbox for name], local advertisement [insert textbox for name], Facebook, Twitter, magazine [insert text box for name of magazine], Other (insert text box)}
10	Please provide your contact information. We will email you to let you know if you are eligible for the study. Preferred method of contact? • Email • Phone d. Name: _________ e. Email address: _______________ f. Phone (optional): ___________	{Checkboxes} {Textboxes}

Scoring Eligibility Criteria for HCPs
1 – n/a (all eligible)
2 – Yes
3 – Yes
4 – n/a (all eligible; unless we block to seek people from specific regions to round out focus groups)
5a – Yes
  5c – yes (but of necessity, they will need to have answered 5a with 'Yes' first)
6 – they must select which disorder group they'd prefer to participate in
7 – Yes
8 – Yes

Appendix E. Parent Focus Group / Interview Topic Guide

Introductory Script

Welcome, everyone. My name is Kim Tan-MacNeill, and I am a clinical psychology PhD student from Dalhousie University in Halifax. This study is part of my dissertation research on sleep problems in children with neurodevelopmental disorders, or NDD, and is supervised by Dr. Isabel Smith and Dr. Penny Corkum. I will be facilitating today's discussion. This is ______[*introduce second moderator*], who will be assisting us today. He/she won't be on screen, but will be available to help you with any technical difficulties and will be helping me take notes.

Thank you for agreeing to participate in our focus group! We appreciate your willingness to share your time and experiences. The information you share with us today will help us to develop effective treatments for children with [ASD / ADHD / FASD / CP] and sleep problems.

What you say here is confidential. We will be recording the video and audio from today's session so that the discussion can be transcribed. When we transcribe the discussion, we will remove all identifying information, such as your names and any other very specific information that might identify you. Once transcribed, we will analyze the discussions for common themes and key ideas that you raise. If you have any questions about this session or the project after the session, feel free to contact me by email or telephone using the contact info provided. Also, if you feel like you didn't get a chance to say something or if there is anything more you want to say after today's session ends, I welcome your emails. I will send you a summary of the study findings after we've finished analyzing the information from these focus groups.

Today, our goal is to talk about your experiences with your children's behavioural sleep problems, called insomnia. I'm especially interested in your experiences with seeking and accessing treatment. Some of you may currently be dealing with your children's sleep problems, while for others, your children may have had sleep problems when they were younger. Some of you may have already sought and tried treatments, while others may not have.

I'll be asking you a series of questions. Sometimes, I'll jump in to move the discussion along, as we have a fair bit to cover and a limited amount of time! If any of you have technical difficulties, please use the chat box at the bottom left of your screen to message [*second moderator]*. If you want to speak and can't get a chance, please click the "raise your hand" button at the bottom left of your screen. As soon as we finish the session today, you will receive a link on your screen taking you to our user feedback survey. Please fill this out – one of our goals is to learn more about conducting online, real-time, audio- and video-based focus groups, as this is a very new way of doing research!

Introductory Question

1. Let's begin by having everyone tell us their first name and how old their child is and whether a boy or girl.

Knowledge of sleep in children with NDD

2. I am interested in hearing from you about your children’s sleep problems, and what (you think) contributes to them.

   Prompts: What types of sleep problem(s) does your child experience?

   What do you think causes them?

For this study, we are specifically interested in your children’s insomnia, which is extremely common in children with [ASD / ADHD / FASD / CP]. The screening questionnaires that you filled out asked whether your children have sleep problems that can be considered insomnia. These include problems following bedtime routines, such as difficulty following parents’ requests and their bedtime schedule; problems falling asleep, such as lying awake in bed for more than 20 minutes after lights out, getting out of bed, and crying at bedtime; problems staying asleep, such as often waking up during the night and having trouble getting back to sleep; and inappropriate early awakenings.

Seeking and Accessing Sleep Treatments

First I’d like to hear about your experiences **seeking** treatment for your children’s **insomnia**. After that we’ll talk about your experiences with different kinds of treatments.

3. What are your thoughts on whether insomnia in children with [ASD / ADHD / FASD / CP] can be treated?
4. Have you ever sought help or treatment for your child’s insomnia?

   Follow-up: From whom/how/where/how did you find out about this?

   Why/what were your reasons for doing so?
5. Once you found or were referred to a treatment, what was your experience with accessing the treatment?

   Prompt: Was it easy or difficult? Were you able to access the treatment? Was this treatment publicly provided, or did you or private insurance have to pay?
6. Thinking back to your experience, what did you find was helpful to you in seeking and accessing treatment?
7. What did you find unhelpful?

Uptake of Sleep Treatments

I’m now interested in your experiences of actually going through treatment for your child’s insomnia. If you haven’t gone through treatment, then I’m curious to hear what you think might be helpful or unhelpful, easy or difficult to do, about the process. There are many different types of treatments, such as prescribed medication, over-the-counter treatments such as melatonin, and behavioural treatments. We’ll talk about them each separately.

8. Let’s briefly talk about medication first. Have any of you tried medication for your child’s insomnia, and how did that work? (If you’ve tried melatonin, we’ll get to that in a minute, but first we’ll discuss prescribed medications.)

   Follow-up: What types of medication?

Was this treatment publicly provided, did you pay with your own money, or use private insurance?
How did you find the process of starting and sticking with the medication? Was it easy / difficult for you and your child?
What was helpful/easy for you and your child?
What was unhelpful/difficult for you and your child?
Thinking back, what would you change, if anything, to better meet the needs of a child with [ASD / ADHD / FASD / CP]?

9. Let's talk about over-the-counter treatments like melatonin and natural remedies. Have any of you tried these for your child's insomnia, and how did that work?

Follow-up: What types of over-the-counter treatments / melatonin?
Was this treatment publicly provided, did you pay with your own money, or use private insurance?
How did you find the process of starting and sticking with the treatment? Was it easy / difficult for you and your child?
What about the treatment was helpful/easy for you and your child?
What was unhelpful/difficult for you and your child?
Thinking back, what would you change, if anything, to better meet the needs of a child with [ASD / ADHD / FASD / CP]?

Now, let's talk about behavioural treatments. These are based on the idea that healthy sleep is a learned behaviour that we can teach kids. Behavioural treatments include parent and child education about sleep, teaching good routines and bedtime habits, and more structured strategies like "extinction," which is also known as the "cry it out approach."."

10. Have you tried any behavioural treatments for your children? Which ones?

Follow-up: Who implemented / conducted the treatment? (e.g., parent or professional)
Was this treatment publicly provided, did you pay with your own money, or use private insurance?

11. How did you find the process of actually trying / starting / implementing the treatment?

Follow-up: Was it easy / difficult for you and your child?
Were you able to stick with it?
How long did you try it for?

12. What about the treatment was helpful/easy for you and your child?

Follow-up: What was unhelpful/difficult for you and your child?
Thinking back, what would you change, if anything, to better meet the needs of a child with [ASD / ADHD / FASD / CP]?

13. Overall, what type of treatment (medication, over-the-counter, behavioural) did you find most effective?
14. What type of treatment did you find least effective?

Acceptability of Treatments

Now, we're going to discuss ways of delivering behavioural treatments for children's sleep problems. Many different methods are used to deliver behavioural treatments. For example, one way might involve meeting weekly with a therapist in a clinic, over a period of 5-8 weeks. The therapist would provide education about sleep and behaviour treatment, and teach you strategies to use with your child, then send you home to work on improving your child's sleep each week.

Another way to deliver treatment might involve in-home support, where a behavioural therapist comes into your home, over-night if necessary, to help you use strategies to improve your child's sleep.

Another method of delivery might be telephone coaching. With this, you would receive a manual containing information about sleep and behaviour treatment, teaching you how to use strategies to improve your child's sleep. You would be able to individualize the strategies to your own child. The manual would have different modules for each week, which you would work through. You would communicate with a coach by telephone to problem solve on a weekly basis.

All behavioural treatments can help parents learn strategies to build better bedtime routines, address specific sleep problems, and improve their children's sleep, among other things.

Our research team is looking at putting these strategies on line, in the form of an interactive web-based intervention for parents of kids with [ASD / ADHD / FASD / CP]. We'd like to find out what parents think about this, especially in comparison to more traditional methods of treatment!

Sample description: Our idea is to give parents access to a website that is specifically designed for parents of children with neurodevelopmental disorders and sleep problems. The website would be organized in modules for each week, with videos and exercises. For example, Week 1 might consist of basic education about sleep in kids with neurodevelopmental disorders. Week 2 might be an introduction to behavioural strategies. Each module teaches how to implement strategies to improve child's sleep. Strategies can be individualized based on child's specific sleep problems/age/etc. Parents enter child's sleep information each week (e.g., sleep diary). The website would be interactive and provides feedback to parents (e.g., how much sleep has changed over past weeks).

Prompt: How would this fit for you and your child?

What are the pros/cons?
What do you think of this compared to the other methods of treatment delivery?

Prompt: If there was an option to have a phone or online coach along with the web intervention, how would this affect your thoughts on whether this is an acceptable treatment or not?

Ending Questions

15. As I mentioned earlier, the information you shared with us today will be used to help inform the development of an effective behavioural treatment for insomnia in children with neurodevelopmental disorders. The goal of this study is to explore barriers and facilitators to parents' access to and use of treatments. Thinking back through our discussion, is there anything we should have talked about but didn't? Have we missed anything? *[pause to allow participants to respond]*

16. Today we discussed sleep in children with [ASD / ADHD / FASD / CP] and treatment for sleep problems, as well as your experiences with seeking and using treatment and some possible types of treatment delivery. What do you see as the main thing that stands in the way of effective sleep treatment for children with [ASD / ADHD / FASD / CP]?

17. Based on your experience, what would you want to tell parents of other children with [ASD / ADHD / FASD / CP] about **seeking, accessing, and using** treatments for insomnia?

    If you feel that there is anything we missed or should have talked about but didn't, or if there's anything that you didn't get a chance to say but wanted to, please feel free to email me.

Concluding Script

I would like to thank everyone for taking the time to participate in our focus group today. You have all contributed to a very interesting discussion and provided a lot of insightful information that will help the study. *[Remind them to fill out the user feedback survey, that the recording will be transcribed/de-identified, that participants will receive a summary of the focus group discussion findings by email, that they'll be entered into a draw to win gift card, and that if they indicated so on the consent form, they may be contacted by us about future research studies.*]

Sample General Prompts:

- Why?
- Tell us more about that.
- Say more about that.
- Could you explain ____ more thoroughly?
- You said, ______; could you tell us more about it?
- How did you feel when that happened?
- What gave you that impression?
- Did anyone else have a similar experience?
- Where did you learn about ________?
- What has been helpful/unhelpful?
- What do you wish other parents of children with [ASD / ADHD/ FASD / CP] and sleep problems knew?
- What would you recommend to other parents?
- What would you change/keep the same?

Appendix F. Health Care Professional Focus Group / Interview Topic Guide

Introductory Script:

Welcome, everyone. My name is Kim Tan-MacNeill, and I am a clinical psychology PhD student from Dalhousie University in Halifax. This study is part of my dissertation research on sleep problems in children with neurodevelopmental disorders and is supervised by Dr. Isabel Smith and Dr. Penny Corkum. I will be facilitating today's discussion. This is ____ [*introduce second moderator*], who will be assisting us today. He/she won't be on screen, but will be available to help you with any technical difficulties and will be helping me take notes.

Thank you for agreeing to participate in our focus group! I know you are all extremely busy, so I appreciate your willingness to share your time and experiences. The information you share with us today will help us to develop effective treatments for children with [ASD / ADHD / FASD / CP], and sleep problems.

What you say here is confidential. We will be recording the video and audio from today's session so that the discussion can be transcribed. When we transcribe the discussion, we will remove all identifying information, such as your names and any other very specific information that may identify you. Once transcribed, we will analyze the discussions for common themes and key ideas that you raise. If you have any questions about this session or the project after the session, feel free to contact me by email or telephone using the contact info provided. Also, if you feel like you didn't get a chance to say something or if there is anything more you want to add after today's session ends, I welcome your emails. I will be sending you a summary of the findings after we've finished analyzing the information from these focus groups.

Our goal today is to discuss your experiences working with children with [ASD / ADHD / FASD / CP], who have behavioural sleep problems. We will focus especially on your experiences with recommending, accessing, and providing treatments for insomnia.

I'll be asking you a series of questions. Sometimes I will jump in to move the discussion along, as we have a fair bit to cover and I value your time! If any of you have technical difficulties, please use the chat box at the bottom left of your screen to message [*second moderator*]. If you want to speak and can't get a chance, please click the "raise your hand" button at the bottom left of your screen and I'll make sure that we hear from you. As soon as we finish the session today, you will immediately receive a link on your screen taking you to our user feedback survey. Please fill this out – one of our goals is to contribute to the literature on conducting online, real-time, audio and video based focus groups, as this is a very new way of doing research!

For our discussion today, I'll be asking you to think about the 4 to 12-year-old children with [ASD / ADHD / FASD / CP], with whom you work, and their families.

Introductory Question

1. Let's begin by having everyone share your name and profession, and tell us about how often you are asked or consulted about sleep problems in kids with [ASD / ADHD / FASD / CP], and what types of sleep problems these are (e.g., behavioural, etc.).

Experience and Involvement with Sleep Treatments

Although kids with [ASD / ADHD / FASD / CP] can have physiologically-based sleep disorders (such as sleep apnea), today we'll focus on behavioural sleep problems, called "insomnia." For our purposes today we will include under this umbrella term the following: bedtime resistance, poor sleep routines and habits, difficulty falling asleep, irregular sleep-wake patterns, and night or early morning awakenings.

I would like to discuss your experiences with recommending or accessing treatments for insomnia.

2. In your regular practice, to what extent are you involved with sleep treatment? (e.g., referring patient/client elsewhere, providing the treatment, etc.)
3. What treatment or interventions do you typically recommend that families pursue OR typically use with families? *(depends on level of HCPs' involvement with treatment)*
4. What experience or familiarity do you have with evidence-based **behavioural interventions** for insomnia for children with [ASD / ADHD / FASD / CP]? Behavioural interventions focus on the idea that healthy sleep is a learned behaviour that we can teach and reinforce. They include components like parent psychoeducation, teaching good sleep habits, and strategies like standard or graduated extinction, scheduled awakenings, faded bedtime, stimulus fading, and chronotherapy.

   Prompt: How often do you recommend behavioural interventions?

Access to Sleep Treatments

5. What kind of access do **you** have to evidence-based treatments for insomnia in kids with [ASD / ADHD / FASD / CP]?

   Follow-up (if not addressed by #2): Do you refer child elsewhere or implement treatment yourself?
6. What factors impact your ability to access treatment?

   Prompt: Family/patient
   Costs to families
   Systems (e.g., hospital, private practice, etc.)
   Education/training

Provision of Treatment

Now, let's discuss your experiences actually providing and implementing these treatments with your patients or clients (for those who do provide treatment).

7. What strategies and treatments were effective and helpful for children with [ASD / ADHD / FASD / CP]?

   Prompt: What worked?
8. What strategies and treatments were not effective or unhelpful?

   Prompt: What didn't work?

9. First, let's talk about medication. Do any of you recommend or prescribe medication for your patients' insomnia?

   Prompt: What types of medication?
   What were your impressions of your patients' (parents/child) experiences with medication?

What about the medication was helpful/easy for your patients?
What about the medication was unhelpful/difficult for your patients?

10. Let's talk about over-the-counter treatments like melatonin and natural remedies. Do any of you recommend over-the-counter treatments?

    Prompt: What types?
    What were your impressions of your patients' (parents/child) experiences with medication?
    What about the treatment was helpful/easy for your patients?
    What about the treatment was unhelpful/difficult for your patients?

11. Let's talk about behavioural treatments. These are based on the idea that healthy sleep is a learned behaviour that we can teach kids. Behavioural treatments include parent and child education about sleep, teaching good routines and bedtime habits, and more structured strategies like "extinction," which is also known as the "cry it out approach." Do any of you recommend or provide behavioural treatments for your patients' insomnia?

    Prompt: Which ones?
    Do you implement/conduct the treatment or teach the parent?
    How did you find the process of providing this treatment?
    What are your impressions of how your patients' (parents/child) experiences with behavioural treatments?
    What about this treatment is helpful/easy for your patients?
    What about this treatment is unhelpful/difficult for your patients?

12. In treatment for sleep problems, do you make any modifications or use certain behavioural strategies specifically for children with [ASD / ADHD / FASD / CP]?

13. *(optional – ask if not addressed by #5/6)* What factors impact your ability to provide treatment?

    Prompt: Family/patient
    Costs to families
    Systems (e.g., hospital system, private practice, etc.)
    Education/training

14. Overall, what type of treatment (medication, over-the-counter, behavioural) did you find most effective?
15. What type of treatment did you find least effective?

Acceptability of Treatments

Now, we're going to discuss different ways of delivering behavioural treatments for children's insomnia. Many methods are used to deliver behavioural treatments. For example, one way might involve parents meeting weekly with a therapist in a clinic, over a period of 5-8 weeks. The therapist would provide education about sleep and behaviour treatment, and teach parents strategies to use with their child, then send them home to work on improving their child's sleep each week.

Another way to deliver treatment might involve in-home support, where a behavioural therapist goes into the home, over-night if necessary, to help parents use strategies to improve their child's sleep.

Another method of delivery might be telephone coaching. With this, parents would receive a manual containing information about sleep and behaviour treatment, teaching them how to use strategies to improve their child's sleep. They would be able to individualize the strategies to their own child. The manual would have different modules for each week, which they would work through. Parents would communicate with a coach by telephone to problem solve on a weekly basis.

All behavioural treatments can help parents learn strategies to build better bedtime routines, address specific sleep problems, and improve their children's sleep, among other things.

Our research team is looking at putting these strategies online, in the form of an interactive web-based intervention for parents of kids with [ASD / ADHD / FASD / CP]. We'd like to find out what health professionals think about this, especially in comparison to more traditional methods of treatment!

Sample description: Our idea is to give parents access to a website that is specifically designed for parents of children with neurodevelopmental disorders, and sleep problems. The website would be organized in modules for each week, with videos and exercises. For example, Week 1 might consist of basic education about sleep in kids with neurodevelopmental disorders. Week 2 might be an introduction to behavioural strategies. Each module teaches how to implement strategies to improve child's sleep. Strategies can be individualized based on child's specific sleep problems/age/etc. Parents enter child's sleep information each week (e.g., sleep diary). The website would be interactive and provides feedback to parents (e.g., how much sleep has changed over past weeks).

Follow-up Questions: How would you feel about referring families to such a treatment?
Do you see yourself as able to deliver such a treatment within your role as a health professional?
What are the pros/cons?
What do you think of this compared to the other methods of treatment delivery?

If there was an option to have a phone or online coach along with the web intervention, how would this affect your thoughts on whether this is an acceptable treatment or not?

Ending Questions

16. As I mentioned earlier, the information you shared with us today will be used to help inform the development of an effective behavioural treatment for insomnia in children with [ASD / ADHD / FASD / CP]. Thinking back through our discussion, is there anything we should have talked about but didn't? Have we missed anything? *[pause to allow participants to respond]*

17. Today we discussed what you know about sleep and [ADHD / FASD / CP] and treatment for sleep problems, as well as your experiences accessing and providing treatment and some possible types of treatment delivery. What do you see as the main thing that stands in the way of effective sleep treatment for children with [ASD / ADHD / FASD / CP]?

Concluding Script

I would like to thank everyone for taking the time to participate in our focus group today. You have all contributed to very interesting discussion and provided a lot of insightful information that will help the study. [*Remind them to fill out the user feedback survey, that the recording will be transcribed/de-identified, that participations will receive a summary of the focus group discussion findings by email, that they'll be entered into a draw to win gift card, and that if they indicated so on the consent form, they may be contacted by us about future research studies.*]

Sample General Prompts:

- Why?
- Tell us more about that.
- Say more about that.
- Could you explain ____ more thoroughly?
- You said, ______; could you tell us more about it?
- How did you feel when that happened?
- What gave you that impression?
- Did anyone else have a similar experience?
- What worked/didn't work? *or* What would/wouldn't work?
- What was helpful/unhelpful?
- What do you wish other HCPs working with children with [ASD / ADHD / FASD / CP], and sleep problems knew?
- What would you recommend?

Appendix G. Copyright Permission for Study 3

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An eHealth insomnia intervention for children with neurodevelopmental disorders: Results of a usability study

Author:
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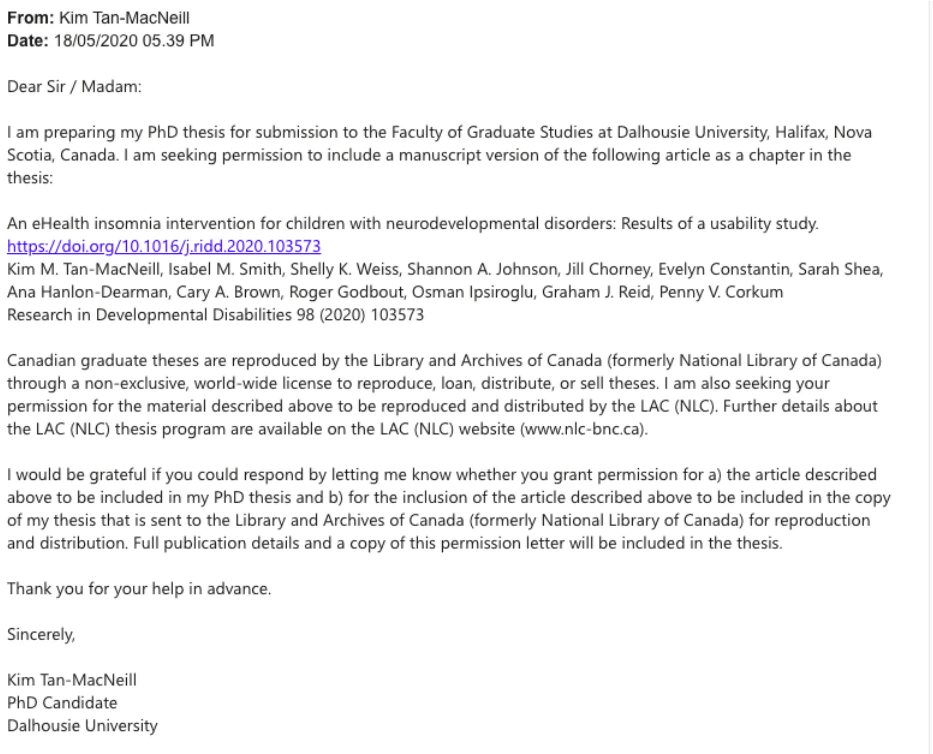

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An eHealth insomnia intervention for children with neurodevelopmental disorders: Results of a usability study.
https://doi.org/10.1016/j.ridd.2020.103573
Kim M. Tan-MacNeill, Isabel M. Smith, Shelly K. Weiss, Shannon A. Johnson, Jill Chorney, Evelyn Constantin, Sarah Shea, Ana Hanlon-Dearman, Cary A. Brown, Roger Godbout, Osman Ipsiroglu, Graham J. Reid, Penny V. Corkum
Research in Developmental Disabilities 98 (2020) 103573

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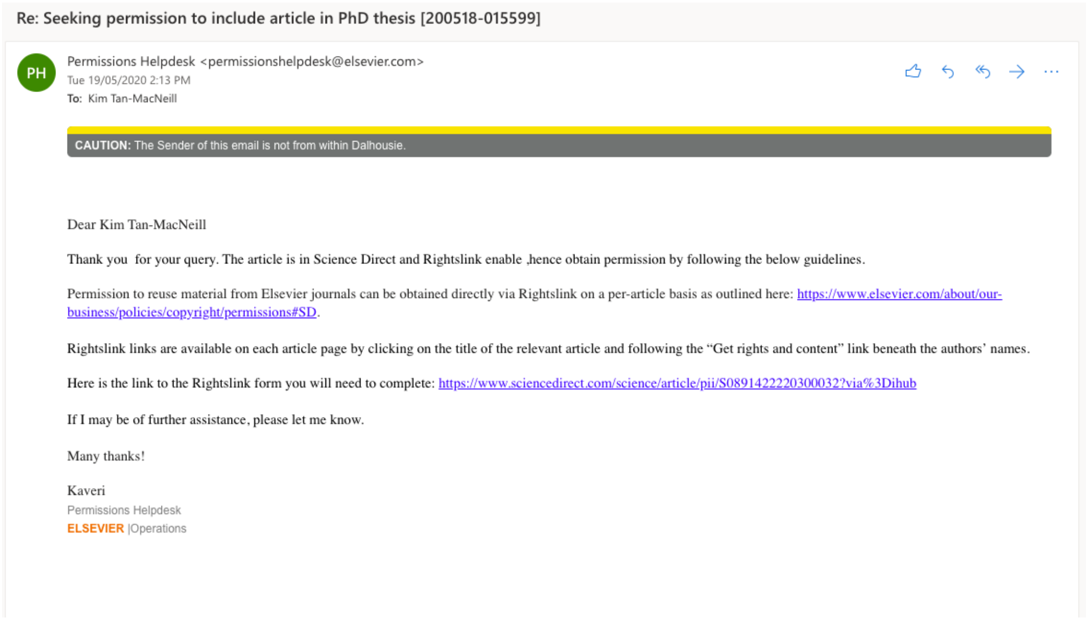
Sincerely,

Kim Tan-MacNeill
PhD Candidate
Dalhousie University



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Appendix H. Study 3 Screening Questionnaire – Modified for NDD (SQ-NDD)

Thank you for agreeing to be screened for the *BNBD: Usability for Children with NDD* study. The first step is to make sure that you and your child meet the criteria to participate in the study. You will be asked to answer 26 questions. This will take about 15 minutes. All questions must be answered in order to complete this section. [All screening questions are mandatory]

1. Are you the legal and primary caregiver of the child who you would like to help through the Better Nights, Better Days program? [Yes/No] [Inclusion criteria - Yes response required]
2. Does your child have one (or more) of the following neurodevelopmental disorders? Select all that apply. [Attention-Deficit / Hyperactivity Disorder (ADHD) / Autism Spectrum Disorder (ASD) / Cerebral Palsy (CP) / Fetal Alcohol Spectrum Disorder (FASD) / No] [Inclusion criteria – Must select at least one ADHD, of ASD, CP, and/or FASD]
3. Do you live in Canada? [Yes/No] [Inclusion criteria - Yes response required]
4. In which province/territory do you live? [Drop down menu of province and territories][Sample distribution factor - Not inclusion / exclusion criteria]
5. What are the first 3 characters of your postal code? [No spaces] [Not inclusion / exclusion criteria]
6. Do you have regular access to a high-speed internet connection? [Yes/No] [Inclusion criteria - Yes response required]
   Technical requirements:
   Operating System: Windows XP or higher or Mac OS X 10.4 or higher
   Browser: Google Chrome 7.0 or higher (recommended), Internet Explorer 8.0 or higher, Firefox 4 or higher, or Safari 5.0 or higher
   Broadband connection: 1 Mbps or higher preferable
7. Are you comfortable communicating in English for day-to-day tasks (e.g., listening to the news on the radio or watching TV, reading books, magazines, etc.)? [Yes/No] [Inclusion criteria - Yes response required]
8. Is your child between 4 and 10 years old (i.e., has not turned 11 yet)? [Yes/No] [Inclusion criteria - Yes response required] [If Yes is clicked, the following question #9 is displayed]
9. What is your child's birth date? [Drop down menu of day, month, year] [Stratification factor]
10. Does your child attend preschool or school? [Yes/No] [Inclusion criteria – Yes response required]
11. Has your child been **diagnosed** with a significant medical disorder (e.g., nighttime asthma attacks, significant or severe nighttime reflux, visual impairment, hearing impairment, tube-feeding) or a medically based sleep disorder (e.g. Obstructive Sleep Apnea, narcolepsy, restless leg syndrome)? [Yes/No] [Exclusion criteria – No response required]

12. Is your child currently being treated with anti-epileptic and/or psychotropic medications (not including stimulant medication), with the medication or dose not stable or expected to change within 6 months? [Yes/No] [Exclusion criteria – No response required]
13. Is your child currently using an over-the-counter (e.g. Benadryl) or natural health medication to treat sleep problems, not including melatonin? [Yes/No] [Exclusion criteria – No response required]
14. Is your child currently using **melatonin** to treat sleep problems? [Yes/No] [Exclusion criteria – response is noted but not used as screening either way – evidence of insomnia symptoms must still be present]
15. Has your child been **diagnosed** with a mental health disorder (including neurodevelopmental disorders, anxiety, depression, psychosis, etc) that **has required or currently requires hospitalization or residential care**? This does not include an Emergency Room visit. [Yes/No] [Exclusion criteria – No response required]
16. Is your child able to walk and independently turn themselves over in bed? [Yes/No] [Exclusion criteria – Yes response required]
17. Is your child able to speak / use verbal language? [Yes/No] [Exclusion criteria – Yes response required]
18. Is your child consistently dry at night (e.g., no wetting bed or bowel incontinence at night)? [Yes/No] [Exclusion criteria – Yes response required]
19. Is your child able to feed him- or herself with a utensil (e.g., fork, spoon)? [Yes/No] [Exclusion criteria – Yes response required]
20. Is your child able to actively participate in dressing him- or herself? [Yes/No] [Exclusion criteria – Yes response required]
21. Is your child able to follow two-step instructions when he or she is focused? (e.g., “First put your shoes on, then get your backpack.”) [Yes/No] [Exclusion criteria – Yes response required]
22. Is your child able to express their preferences using verbal language? [Yes/No] [Exclusion criteria – Yes response required]
23. Is your child able to speak in sentences that contain **at least four words**? (e.g., “I want a dog,” “Give me more milk,” “I need help please”) [Yes/No] [Exclusion criteria – Yes response required]
24. Does your child have one of the following sleep problems?
    a. Does your child have problems falling asleep?
       Problems falling asleep means lying awake in bed after lights out for an extended period of time (20 minutes or more). Sometimes children will call out to parents, or get out of bed. Some children may also cry at bedtime. [Yes/No]
    b. Does your child have problems following bedtime routines?
       Problems following bedtime routines means resistance to parents’ requests, resistance following a schedule, needing constant reminders for each task during the bedtime routine, etc. [Yes/No]
    c. Does your child require you or another adult to be present in order to fall asleep?

Select "Yes" only if you believe this is a problem for you, your child, and/or your family. [Yes/No]

[Inclusion criteria - Yes response required for any of the following: 10 a, b, or c]

25. The Better Nights, Better Days sleep program has been developed for those parents whose child sleeps independently from them (i.e., in a separate room) or those parents who would **like** their child to sleep independently from them. It has not been designed for parents who want to sleep in the same bed or bedroom as their child. Which of the following best describes where you believe your child **SHOULD** sleep? (Choose one) [Exclusion criteria - Must select a, b, d, NOT c]
    a. I intentionally sleep separately from my child because I believe that is the best arrangement.
    b. I prefer to sleep separately from my child, but I do sleep with him/her because my child can't or won't sleep well apart from me
    c. I intentionally sleep with my child because I believe that is the best arrangement
    d. I sleep in the same bed with my child because this is the only option given our space limitations
26. How did you hear about this study and Better Nights, Better Days? (Check all that apply) [Not inclusion / exclusion criteria]
    a. Google search
    b. Family doctor
    c. Pediatrician
    d. Psychologist
    e. Other healthcare provider
    f. Family member or friend
    g. Colleague
    h. Mailing List [If selected, text box appears for name/organization]
    i. Advertisement (e.g., newspaper, newsletter, notice at your child's school or doctor's office etc.) [If selected, text box appears:]
    j. Facebook
    k. Twitter
    l. Magazine [If selected, textbox appears for name of magazine]
    m. Website [If selected, textbox appears for name/address of website]
    n. Other [if selected, textbox appears to specify source]

Appendix I. Study 3 Health-Related Questionnaire-Modified (HRQ-M)

The following questions ask about your child's neurodevelopmental disorder diagnosis, and any other diagnoses or conditions that they may have (including other neurodevelopmental disorders, mental health or health conditions and sleep disorders). These questions are about long-term diagnoses or conditions that your child may be experiencing now or has experienced in the past. A long-term condition is one that has lasted or is expected to last for 6 months or longer and has been diagnosed by a professional such as a family physician, paediatrician, or psychologist.

The questionnaire is broken into these parts:

1. Your Child's Primary Diagnosis (e.g., their ADHD, ASD, CP, or FASD)

2. Any Other Diagnoses
   a) Other neurodevelopmental disorder [NDD] diagnoses or conditions (e.g., ADHD, ASD, CP, or FASD)
   b) Physical health diagnoses or conditions
   c) Mental health diagnoses or conditions
   d) Sleep disorders or conditions

** We know that NDDs often occur together. If your child has more than one NDD please do the following:

- If your child has ASD and another NDD (but not FASD), fill out ASD as the Primary Diagnosis and put the other NDD(s) in the 'Any Other Diagnosis' section

- If your child has FASD and another NDD (including ASD), fill out **FASD** as the Primary Diagnosis and put the other NDD(s) (including ASD) in the 'Any Other Diagnosis' section

This questionnaire will take about 8 minutes to complete.

When answering these questions please consider what your child's behaviour is usually like over the **past month**. If your child's behaviour has been unusual for a specific reason (e.g., sickness, away on vacation) during this time, choose the most appropriate answer that captures your child's typical behaviour.

PART 1: YOUR CHILD'S PRIMARY DIAGNOSIS

1. What primary neurodevelopmental disorder is your child currently diagnosed with? If your child has more than one disorder, remember that the primary diagnosis will be ASD or FASD. [Multiple choice: ADHD, ASD, CP, FASD] [Mandatory to fill out]
2. When was the diagnosis made? (Please select the month and year) [Drop down for month and year]
3. Who diagnosed your child? {Please check all that apply}
   a. Psychologist
   b. Psychiatrist
   c. Family physician

   d. Pediatrician
   e. Other [If selected, text box appears]

4. Is your child currently taking any medications for this? [Yes/No] [If Yes, question 5-7 appear]
   5. Name of the medication [Textbox]
   6. Dose of the medication (i.e. 10mg) [Textbox]
   7. How many times a day is the medication given? [Radio buttons: 1, 2, 3, 4, 5 or more]

PART 2: ANY OTHER DIAGNOSES

Other Neurodevelopmental Disorders

1. Does your child have any other neurodevelopmental disorder diagnoses, in addition to the primary diagnosis that you already described (e.g., ADHD, ASD, CP, FASD)? [Yes/No] [If yes, questions 2-6] appear; if No, skips to **Physical Health Conditions and Disorders**]
2. How many additional neurodevelopmental disorders are diagnosed, **not including** the primary diagnosis you already described? [Radio buttons: 1, 2, 3, 4, 5 or more]

3. What is the diagnosis (e.g., ASD, ADHD, FASD, CP)? [Multiple choice: ADHD, ASD, CP, FASD, Other – textbox for other] [Repeated for each diagnosis indicated in Q2]

4. When was the diagnosis made? (Please select the month and year) [Drop down for month and year]
5. Who diagnosed your child? {Please check all that apply}
   a. Psychologist
   b. Psychiatrist
   c. Family physician
   d. Pediatrician
   e. Other [If selected, text box appears]
6. Is your child currently taking any medications for this? [Yes/No] [If Yes, question 5-7 appear]
   7. Name of the medication [Textbox]
   8. Dose of the medication (i.e. 10mg) [Textbox]
   9. How many times a day is the medication given? [Radio buttons: 1, 2, 3, 4, 5 or more]

Physical Health Conditions and Disorders

1. Is your child diagnosed with a long-term (6 months or more) physical health/medical condition, **not including** CP? (e.g., epilepsy, asthma, celiac disease) [Yes/No] [If No, the parent is automatically directed to **Mental Health Conditions and Disorders**]
2. How many conditions are diagnosed? [Radio buttons: 1, 2, 3, 4, 5 or more]
3. What is the diagnosis? [Repeated for each diagnosis indicated in Q2]

4. When was the diagnosis made? (Please select the month and year) [Drop down for month and year] [Repeated for each diagnosis indicated in Q2]
5. Who diagnosed your child? (Please check all that apply) [Repeated for each diagnosis indicated in Q2]
    a. Family physician
    b. Pediatrician
    c. Other [If selected, text box appears]
6. Is your child currently taking any medications for this? [Yes/No] [If Yes, question 7-9 appear]
7. Name of the medication [Textbox]
8. Dose of the medication (i.e. 10mg) [Textbox]
9. How many times a day is the medication given? [Radio buttons: 1, 2, 3, 4, 5 or more]

Mental Health Conditions and Disorders

1. Is your child diagnosed with a current mental health condition or disorder? (e.g. anxiety, depression, phobia, bipolar disorder, psychosis, obsessive compulsive disorder) [Yes/No] [If No, the parent is automatically directed to **Sleep Disorders**]
2. How many conditions are diagnosed? [Radio buttons: 1, 2, 3, 4, 5 or more]
3. What is the diagnosis? [Repeated for each diagnosis indicated in Q2]
4. When was the diagnosis made? (Please select the month and year) [Drop down for month and year] [Repeated for each diagnosis indicated in Q2]
5. Who diagnosed your child? {Please check all that apply} [Repeated for each diagnosis indicated in Q2]
    a. Psychologist
    b. Psychiatrist
    c. Family physician
    d. Pediatrician
    e. Other [If selected, text box appears]
6. Is your child currently taking any medications for this? [Yes/No] [If Yes, question 7-9 appear]
7. Name of the medication [Textbox]
8. Dose of the medication (i.e. 10mg) [Textbox]
9. How many times a day is the medication given? [Radio buttons: 1, 2, 3, 4, 5 or more]

Sleep Conditions and Disorders

1. Does your child currently have a **sleep disorder** that has been diagnosed by a professional? (e.g., sleep apnea, narcolepsy, restless leg syndrome) [Yes/No] [If No, the parent is automatically directed to **the next Eligibility Questionnaire, the SILS**]
2. How many conditions are diagnosed? [Radio buttons: 1, 2, 3, 4, 5 or more]
3. What is the diagnosis? [Repeated for each diagnosis indicated in Q2]
4. When was the diagnosis made? (Please select the month and year) [Drop down for month and year] [Repeated for each diagnosis indicated in Q2]

5. Who diagnosed your child? {Please check all that apply} [Repeated for each diagnosis indicated in Q2]
    a. Family physician
    b. Pediatrician
    c. Psychologist
    d. Psychiatrist
    e. Sleep specialist
    f. Other [If selected, text box appears]
6. Is your child currently taking any medications for this? [Yes/No] [If Yes, question 7-9 appear]
7. Name of the medication [Textbox]
8. Dose of the medication (i.e. 10mg) [Textbox]
9. How many times a day is the medication given? [Radio buttons: 1, 2, 3, 4, 5 or more]

Appendix J. Study 3 End-of-Session Usability Questionnaire

This appendix contains the End-of-Session Usability Questionnaire that was administered to participants after each of the five sessions (five times in total), through an Opinio questionnaire that was emailed to the participants when they completed each session. Questions 20 and 21 were excluded from the Session 1 questionnaire (since parents had not completed homework yet). After Session 5, they also completed the "Overall Usability Questionnaire" (see Appendix 7 for this questionnaire).

Legend:
RED: Internal instructions for programming (Opinio) or the Research Coordinator
GREEN: Participant response options
BLUE TEXTBOX: In-house Introduction

End-of-Session Usability Questionnaire

The following questions are about your impressions of Session [1/2/3/4/5] of *Better Nights, Better Days (BNBD)*. We will use the information you provide to help us as we develop a version of *BNBD* for children with neurodevelopmental disorders. As such, please provide comprehensive and candid responses based on your perception of the program.

You will be asked to read and rate a series of statements, and provide comments that explain your ratings.

Please enter your Participant ID: [textbox]

Date Completed (mm.dd.yyyy): [automatically captured by Opinio

Useful

1. This session provided information that helped me to better understand, and would help me treat, my child's insomnia.
   - ○ Strongly agree
   - ○ Agree
   - ○ Neither agree nor disagree
   - ○ Disagree
   - ○ Strongly disagree
2. This session provided information that was useful to me as a parent of a child with a neurodevelopmental disorder who has insomnia.
   - ○ Strongly agree
   - ○ Agree
   - ○ Neither agree nor disagree
   - ○ Disagree
   - ○ Strongly disagree
3. Please provide comments that support your ratings about the **usefulness** of this session. Include any suggestions you may have to improve **usefulness:** *[textbox]*

Usable/Findable

4. This session was user-friendly and could be navigated with ease.
   - ○ Strongly agree
   - ○ Agree
   - ○ Neither agree nor disagree

  - ○ Disagree
  - ○ Strongly disagree
5. This session took a reasonable amount of time to complete.
  - ○ Strongly agree
  - ○ Agree
  - ○ Neither agree not disagree
  - ○ Disagree
  - ○ Strongly disagree
6. Please provide comments that support your rating about the **usability/findability** of this session. Include any suggestions you may have to improve **usability/findability**: *[textbox]*

Desirable

7. This session was visually appealing and the organization of information on the screen was clear. The *way* the information was presented (e.g., design, colour, font, graphics) was a positive addition to my user experience.
  - ○ Strongly agree
  - ○ Agree
  - ○ Neither agree nor disagree
  - ○ Disagree
  - ○ Strongly disagree
8. Please provide comments that support your rating about the **desirability** of this session. Include any suggestions you may have to improve **desirability**: *[textbox]*

Valuable

9. Overall, the information provided in this session is valuable to me.
  - ○ Strongly agree
  - ○ Agree
  - ○ Neither agree nor disagree
  - ○ Disagree
  - ○ Strongly disagree
10. Please provide comments that support your rating about the **value** of this session. Include any suggestion you may have to improve the **value** of this session: *[textbox]*

Accessible

11. I was able to access the session from the device(s) I chose to use at a time and location that was convenient for me.
  - ○ Strongly agree
  - ○ Agree
  - ○ Neither agree nor disagree
  - ○ Disagree
  - ○ Strongly disagree
12. The information provided in this session was easy for me to understand.
  - ○ Strongly agree
  - ○ Agree
  - ○ Neither agree nor disagree
  - ○ Disagree
  - ○ Strongly disagree
13. Please provide comments that support your rating about the **accessibility** of this session. Include any suggestion you may have to improve the **accessibility** of this session: *[textbox]*

Credible

14. Overall, I believe that the information provided in this session comes from a reputable source, and I trust the information enough to feel comfortable using it to treat my child's insomnia.
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree
15. Please provide comments that support your rating about the **credibility** of this session. Include any suggestions you may have that would help make the information appear more **credible**: *[textbox]*

Acceptable

16. The information provided in this session was acceptable to me, as a parent of a child with a neurodevelopmental disorder and insomnia.
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree
17. Please provide comments that support your rating about the **acceptability** of this session. Include any suggestions that you may have to improve the **acceptability** of this session: *[textbox]*

Videos

18. The videos in this session helped me learn the material..
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree
19. Please provide comments that support your rating about the **videos** in this session. Include any suggestions you may have to improve the **videos** included in the session: *[textbox]*

Activities

20. The activities within the session are helpful, and I would refer to them/use them in future.
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree
21. Please provide comments that support your rating about the **activities** within this session. Include any suggestions you may have to improve the **activities** included in the session: *[textbox]*

Homework (in Session Plan) [excluded from Session 1 questionnaire – branching]

22. The homework included in this session's session plan (e.g., reminders, goals, and activities to do at home) was helpful and manageable.
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree

23. Please provide comments that support your rating about the **homework** in this session. Include any suggestions you may have about the **homework** in this session: *[textbox]*

Features (e.g., Printable Worksheets, Summary Sheets, Supplemental Materials)

24. The features in this session (including printable worksheets, summary sheets and any supplemental materials) are helpful, and I would refer to them in future.
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree
25. Please provide comments that support your rating about the **features** of this session. Include any suggestions you may have to improve the **features** included in this session: [*textbox]*

Support

26. Overall, I felt like I had adequate support, clarification, and motivation while completing this session.
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree
27. Please provide comments that support your rating about the **support** in this session. Include any suggestions you may have to improve **support** in this session: *[textbox]*

Feasibility

28. Overall, I was able to fully participate in this session by completing activities and using or implementing the strategies with my child without any major obstacles.
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree
29. Please provide comments that support your rating about the **feasibility** of this session. Include any suggestions you may have to improve the **feasibility** of this session or describe anything that made the session more or less **feasible**: *[textbox*]

Readiness

30. How ready is this session for use with parents of children with the same neurodevelopmental disorder as your child and insomnia?
    - ○ Extremely ready
    - ○ Very ready
    - ○ Moderately ready
    - ○ Slightly ready
    - ○ Not at all ready

General Comments

31. Please provide any additional feedback you have about the session. If you do not believe it is **ready for use**, in what ways must it be modified to be ready? *[textbox]*

Appendix K. Study 3 Overall Usability Questionnaire

This appendix contains the Overall Usability Questionnaire that was administered to participants after they completed the entire intervention. It was completed immediately after the Session 5 End-of-Session Usability Questionnaire. A link was emailed to participants.

Legend:
RED: Internal instructions for programming (Opinio) or the Research Coordinator
GREEN: Participant response options
BLUE TEXTBOX: In-house Introduction

Overall Usability Questionnaire

The following questions are about your impressions of the *Better Nights, Better Days (BNBD)* intervention *overall*. We will use your responses to help us develop a version of *BNBD* for children with neurodevelopmental disorders. As such, please provide comprehensive and candid responses based on your perception of the program.

Please enter your Participant ID: [textbox]

Date Completed (mm.dd.yyyy): [automatically captured by Opinio

Technology, Hardware, and Website

The following questions are about the technology and hardware you used as you went through the BNBD intervention and website.

1. What type of hardware did you typically use? You can select multiple options.
   - ○ Desktop computer
   - ○ Laptop computer
   - ○ Tablet
   - ○ Smartphone
   - ○ Other [text box]

2. What Operating System (OS) did you typically use? You can select multiple options.
   - ○ Windows
   - ○ Mac OS
   - ○ iOS (Apple smartphones)
   - ○ Google/Android (specific to smartphones)
   - ○ Other [textbox]

   What version of the OS did you use? [textbox]

3. What internet browser did you typically use? You can select multiple options.
   - ○ Firefox
   - ○ Safari
   - ○ Google Chrome
   - ○ Internet Explorer
   - ○ Edge
   - ○ Other [textbox]

   What version of the browser did you use? [textbox]

4. How quickly did the BNBD site typically load on the device you used most often?
   - ○ 1 second

- 2 seconds
- 3 seconds
- 4 seconds
- 5 seconds
- More than 5 seconds

5. How often did the site crash?
    - Never
    - 1-2 times
    - 3-4 times
    - 4-5 times
    - More than 5 times

If it did crash, what was happening when it did? [textbox]

The following statement pertains to your impression of the BNBD homepage.

6. The BNBD dashboard page (the page that appears when you log in) adequately highlights the features of the intervention.
    - Strongly agree
    - Agree
    - Neither agree nor disagree
    - Disagree
    - Strongly disagree
7. Please provide comments that support your rating of the **homepage** and include any suggestions you may have to improve the **homepage**: [textbox]

Overall User Experience and Impressions

The following statements pertain to your impressions of the BNBD intervention overall. Please indicate the extent to which you agree with the following statements.

Useful

8. Overall, this intervention provided information that helped me to better understand, and would help me treat, my child's insomnia.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
9. Overall, this intervention provided information that was useful to me as a parent of a child with a neurodevelopmental disorder who has insomnia.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
10. Please provide comments that support your ratings about the **usefulness** of this intervention. Include any suggestions you may have to improve **usefulness:** *[textbox]*

Usable/Findable

11. Overall, this intervention was user-friendly and could be navigated with ease.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree

    e. Strongly disagree

12. Overall, this intervention took a reasonable amount of time to complete.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
13. Please provide comments that support your rating about the **usability/findability** of this intervention. Include any suggestions you may have to improve **usability/findability**: *[textbox]*

Desirable

14. Overall, this intervention was visually appealing and the organization of information on the screen was clear. The *way* the information was presented (e.g., design, colour, font, graphics) was a positive addition to my user experience.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
15. Please provide comments that support your rating about the **desirability** of this intervention. Include any suggestions you may have to improve **desirability**: *[textbox]*

Valuable

16. Overall, the information provided in this intervention is valuable to me.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
17. Please provide comments that support your rating about the **value** of this intervention. Include any suggestion you may have to improve the **value** of the intervention: *[textbox]*

Accessible

18. Overall, I was able to access the intervention from the device(s) I chose to use at a time and location that was convenient for me.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
19. Overall, the information provided in the intervention was easy for me to understand.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
20. Please provide comments that support your rating about the **accessibility** of the intervention. Include any suggestion you may have to improve the **accessibility** of the intervention: *[textbox]*

Credible

21. Overall, I believe that the information provided in this intervention comes from a reputable source, and I trust the information enough to feel comfortable using it to treat my child's insomnia.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
22. Please provide comments that support your rating about the **credibility** of this intervention. Include any suggestions you may have that would help make the information appear more **credible**: *[textbox]*

Acceptable

23. The information provided in this intervention was acceptable to me, as a parent of a child with a neurodevelopmental disorder and insomnia.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
24. Please provide comments that support your rating about the **acceptability** of this intervention. Include any suggestions that you may have to improve the **acceptability** of this intervention: *[textbox]*

Videos

25. Overall, I found the videos (not including the Expert Videos) in this intervention helpful.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
26. Overall, I found the Expert Videos in this intervention helpful.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
27. Overall, I enjoyed the videos presented in this intervention.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
28. Overall, the videos added educational value to the intervention.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
29. Please provide comments that support your rating about the **videos** in this session. Include any suggestions you may have to improve the **videos** included in the intervention: *[textbox]*

Activities

30. Overall, I found the activities within each session in the intervention helpful.
    a. Strongly agree

    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree

31. Overall, I found the activities within each session in the intervention enjoyable.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
32. Please provide comments that support your rating about the within-session **activities** in this intervention. Include any suggestions you may have to improve the **activities** included in the intervention: *[textbox]*

Homework (in Session Plan)

33. Overall, I found the homework included in each session's session plan (e.g., reminders, goals, and activities to do at home) throughout the intervention helpful and manageable.
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree
34. Please provide comments that support your rating about the **homework** in this session. Include any suggestions you may have about the **homework** in the intervention: *[textbox]*

Support

35. Overall, I felt like I had adequate support, clarification, and motivation while completing the intervention
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree
36. Please provide comments that support your rating about the **support** in the intervention. Include any suggestions you may have to improve **support** in the intervention: *[textbox]*

Feasibility

37. Overall, I was able to fully participate in the intervention by completing activities and using or implementing the strategies with my child without any major obstacles.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
38. Please provide comments that support your rating about the **feasibility** of this session. Include any suggestions you may have to improve the **feasibility** of the intervention or describe anything that made the intervention more or less **feasible**: *[textbox]*

Key Features

The following items pertain to your impressions of the helpfulness and importance of each of the key features of BNBD.

39. The **Sleep Diary** was a helpful and important feature of the intervention.
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree

40. The **Session Summaries** were helpful and important features of the intervention.
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree
    - ○ N/A – Did not access. Please explain why you did not access the Session Summaries: *[textbox]*

41. The **Session Plans** were helpful and important features of the intervention.
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree
    - ○ N/A – Did not access. Please explain why you did not access the Printable Session Plans: *[textbox]*

42. The **Roadblocks documents** were a helpful and important feature of the intervention.
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree
    - ○ N/A – Did not access. Please explain why you did not access the “Roadblocks” section: *[textbox]*

43. The **“My Stuff” section** was a helpful and important feature of the intervention.
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree
    - ○ N/A – Did not access. Please explain why you did not access the “My Stuff” section: *[textbox]*

44. The ability to **“Review Session**” was a helpful and important feature of the intervention.
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree
    - ○ Disagree
    - ○ Strongly disagree
    - ○ N/A – Did not access. Please explain why you did not access the “Review Session” feature: *[textbox]*

45. The “**Reward Centre”** was a helpful and important feature of the intervention.
    - ○ Strongly agree
    - ○ Agree
    - ○ Neither agree nor disagree

- Disagree
- Strongly disagree
- N/A – Did not access. Please explain why you did not access the “Reward Centre”: *[textbox]*

46. The **Homework Check-In and Feedback** were helpful and important features of the intervention.
    - Strongly agree
    - Agree
    - Neither agree nor disagree
    - Disagree
    - Strongly disagree
    - N/A – Did not access. Please explain why you did not access the Homework Check-In and Feedback: *[textbox]*

47. The **“How Are Things Going?” Section and Feedback** were helpful and important features of the intervention.
    - Strongly agree
    - Agree
    - Neither agree nor disagree
    - Disagree
    - Strongly disagree
    - N/A – Did not access. Please explain why you did not access the “How Are Things Going?” Section and Feedback: *[textbox]*

48. The **“Looking Ahead” section** was a helpful and important feature of the intervention.
    - Strongly agree
    - Agree
    - Neither agree nor disagree
    - Disagree
    - Strongly disagree
    - N/A – Did not access. Please explain why you did not access the “Looking Ahead” section: *[textbox]*

49. The “**How to Use” tutorial section** was a helpful and important feature of the intervention.
    - Strongly agree
    - Agree
    - Neither agree nor disagree
    - Disagree
    - Strongly disagree
    - N/A – Did not access. Please explain why you did not access the “How to Use” section: *[textbox]*

50. Overall, I was able to download and access the features of this intervention.
    - Strongly agree
    - Agree
    - Neither agree nor disagree
    - Disagree
    - Strongly disagree

51. Please provide comments that support your ratings about the **key features/sections** of the intervention, including any suggestions you may have to improve them: *[textbox]*

52. The **automatic feedback on my child’s progress** provided throughout was a helpful and important feature of the intervention.
    - Strongly agree

- Agree
- Neither agree nor disagree
- Disagree
- Strongly disagree

53. The **mobile device friendliness** was a helpful and important feature of the intervention.
    - Strongly agree
    - Agree
    - Neither agree nor disagree
    - Disagree
    - Strongly disagree
    - N/A – Did not use a mobile device.

54. The **ability to take part in the intervention by distance** (e.g., at home, rather than in a clinic / doctor's office, etc.) was a helpful and important feature of the intervention.
    - Strongly agree
    - Agree
    - Neither agree nor disagree
    - Disagree
    - Strongly disagree

Video Testimonials

We plan to include parent testimonial videos about their experiences with BNBD throughout the intervention. Please rate the following statement:

55. Parent video testimonials would enhance the *BNBD* intervention.
    - Strongly agree
    - Agree
    - Neither agree nor disagree
    - Disagree
    - Strongly disagree

56. Please provide comments that support your rating of **parent video testimonials** and include any suggestions you may have: *[textbox*]

57. Would you be willing to provide a parent video testimonial? If you are interested in hearing more about this, please click yes, and you will be contacted at a later date by the research team.
    a. Yes
    b. No

General Comments and Feedback

58. Everything that I expected and hoped to see in this intervention was present.
    a. Strongly agree
    b. Agree
    c. Neither agree nor disagree
    d. Disagree
    e. Strongly disagree
59. Overall, how satisfied were you with this intervention?
    a. Very satisfied
    b. Satisfied
    c. Neither satisfied nor dissatisfied
    d. Dissatisfied
    e. Very dissatisfied

60. Overall, how ready do you think this intervention is for use with parents of children with the same neurodevelopmental disorder as your child and insomnia?
    a. Extremely ready
    b. Very ready
    c. Moderately ready
    d. Slightly ready
    e. Not at all ready
61. Would you recommend this intervention to other parents of children with the same neurodevelopmental disorder as your child and insomnia whom you know?
    a. Yes
    b. Maybe
    c. No
62. Please provide comments that support your above ratings of the intervention overall, including any suggestions to improve the intervention overall: [*textbox*]

Thank you!

Appendix L. Usability Study: How to Manual for Parents

Usability Study:
How to Manual for Parents

Better Nights
Better Days
for children with
Neurodevelopmental
disorders

Welcome!

Thank you for taking part in the **Better Nights, Better Days: Usability for NDD** study!

As you know from reading through the consent and information forms, this is a **usability** study. This means which means that you will be not only using and doing the BNBD intervention with your child, but also providing us with feedback on your experience of using it. It will be important for you to keep that in mind as you go through. We are most interested in hearing from you:

- what you think worked
- what didn't work
- what was hard
- what was easy
- what you think might need to be changed.

One thing that is different about this study is we are studying how usable and manageable the BNBD intervention is for kids with neurodevelopmental disorders, rather than its effectiveness. As you know, BNBD was originally developed for typically developing children, and its effectiveness with them is currently being studied in another research study. This usability study that you are part of is about learning how we may need to modify BNBD to make it appropriate for families of kids with neurodevelopmental disorders.

Starting the Study

You will receive an email from **bnbd.participate@dal.ca** with a link to the BNBD intervention and instructions on how to log in (username / password).

Study Contact Information and Emails

You'll notice that you will receive emails from two different addresses:

1) sleepndd@dal.ca = the Usability Study (usually Kim Tan-MacNeill)

> Please make sure to always read the emails from sleepndd@dal.ca! This will usually be Kim Tan-MacNeill (lead investigator) emailing you with updates and links to the usability questionnaires that you will be filling out.

2) bnbd.participate@dal.ca = the BNBD intervention

The BNBD intervention is automatically programmed to send you reminder emails! You will receive reminders to complete each session, as well as daily reminders to complete sleep diaries. All of these emails will come from bnbd.participate@dal.ca . Please disregard any references to the BNBD randomized controlled trial (RCT) study or having eight months to complete the study – these are for participants in the ongoing BNBD-for-typically-developing-kids study!

If you have any questions, your best bet is to email Kim at **sleepndd@dal.ca** ! However, if they are specifically related to the intervention or the technical side of things (e.g., how to fill out a questionnaire, something related to the website function itself), you could also email **bnbd.participate@dal.ca** -- make sure you mention that you are part of the **Usability** study.

Completing Session and Sleep Diaries

There are 5 sessions in total. You will have up to 2 weeks to complete each session. It takes between 1-2 hours to complete a session start to finish, which involves watching some videos, completing interactive activities, and doing some reading. Some people like to stop and come back later (you can log back in; BNBD will remember where you left off).

One tip: you have to watch videos all the way to the end before the website will allow you to click “Next” to the next page of the intervention!

As soon as you finish a session, a 7-day waiting period begins. During this time, you have to wait the full 7 days AND complete 5 sleep diaries for your child before you have access to the next session. The 5 sleep diaries need to be completed within a 14-day time span. So basically you have a minimum of 1 week and up to 2 weeks to get those sleep diaries filled out before you can move on to the next session!

If you don’t complete 5 sleep diaries within a 14-day period, the “sleep diary clock” resets, and you’ll need to do 5 diaries within a new 14-day period.

There are instructions in the intervention itself about how to fill out the sleep diaries, and as I mentioned, the intervention itself will send you reminders if you haven’t completed 5 yet. If you have any questions, please email us and ask!

Completing the Usability Questionnaires After Each Session

One week after you’ve completed a session (whether or not you’ve done the sleep diaries yet), we will email you a link to the Usability Questionnaire for that session. It is hosted on a separate website. Please try out the session strategies and activities with your child *before* filling out the Usability Questionnaire! It’s important for you to have a sense of how they went. The Usability Questionnaires will each take about 20 minutes and you’ll have a chance to share your opinions and suggestions.

Coming to the End of the Study

Completing the five intervention sessions should take you between 10 – 12 weeks. After the fifth and final session, we'll send you a link to complete an Overall Usability Questionnaire about the whole intervention.

After that, we'll schedule a short "exit interview" with you (which we can do online with video conferencing software or by phone). At that point, we'll also ask you if you're interested in providing a video testimonial about your experience with the BNBD intervention.

Compensation!

As a thank you for your participation, time, and efforts, you have the opportunity to receive up to a $100 gift card in compensation. You will receive a $15 gift card for each of the six usability questionnaires you complete (the five session questionnaires and the one overall questionnaire) as well as a $10 gift card for participating in the exit interview. If you withdraw from the study early, you will be compensated for the questionnaires that you have completed.

If you have any questions, please don't hesitate to email us at sleepndd@dal.ca or phone Kim at ###-###-#### (or email her and she can phone you).