



Review Article

Efficacy of parent training incorporated in behavioral sleep interventions for children with autism spectrum disorder and/or intellectual disabilities: a systematic review

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ABSTRACT

Objective: Behavioral sleep interventions are regularly used to improve sleep problems experienced by children with autism spectrum disorder (ASD) and/or intellectual disability (ID). Recent developments have seen the introduction of parent sleep education and healthy sleep practice training to sleep interventions. This article aims to systematically review the evidence on the efficacy of parent training that is incorporated within recent sleep interventions for children with ASD and/or ID.

Method: Electronic databases and manual searches of reference lists identified 11 studies ($n = 416$ children) that met the inclusion criteria.

Results: The evidence presented in this systematic review would suggest that the inclusion of parent training within behavioral sleep interventions for children with ASD and/or ID is generally effective and valued by parents. Nine of the 11 studies reviewed reported a reduction in sleep problems.

Conclusion: The literature conveys an emerging evidence-based practice that could contribute to future behavioral sleep research and guide best-practice decisions to support effective parent training to improve sleep outcomes for children with ASD and/or ID.

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1. Introduction

Autism spectrum disorder (ASD) is defined as a pervasive neurodevelopmental disorder which can be characterized by a triad of persistent impairments with core deficits in social interaction, language and communication, as well as restrictive, repetitive thoughts, routines and behavior patterns [4]. Individuals with ASD can often experience several comorbid medical conditions including sleep disorders [46]. An intellectual disability (ID) is defined as a disability which is characterised by an intellectual impairment that may significantly impede mental capacity such as learning, reasoning, understanding which can affect social functioning and practical skills [3]. Previous research has suggested there may be an overlap with intellectual disabilities and those diagnosed with ASD [27]. However, this may not always be the case, as researchers have found that due to the broad and varied nature of the spectrum of autism, some individuals with ASD may not have impaired intellectual functioning

[20]. Noterdaeme and Euders [35] found that individuals with ASD can be diagnosed with or without an ID. It is evident from previous research that a person can have a diagnosis of ASD and/or ID. Considering that several sleep studies include children with ASD and/or ID, sleep interventions within the current context were reviewed with a potential comorbidity between ASD and/or ID.

Sleep is an essential aspect of life, and a high percentage of children with ASD [26] and ID [40] experience sleep problems. It is reported that 40–80% of children with ASD and/or ID (ASD/ID) present with sleep disturbances [38] and that 58% of children with mild to profound ID experience sleep problems [40]. The abbreviated term “ASD/ID” used hereafter represents children with either a single diagnosis of ASD or ID or a combined diagnosis of ASD and ID. Within the context of the current review, the terms ‘sleep difficulties’, ‘sleep problems’, and ‘sleep disturbance’ are interchangeable and represent the sleep difficulties commonly reported by parents of children with ASD/ID. These include adhering to bedtime routines, sleep initiation, frequent nighttime waking, early morning waking, and co-sleeping with parents. Sleep disturbances are distressing for the child and their families and often result in reduced daily functioning [25]. Sleep

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deprivation can exacerbate the symptoms of ASD/ID, which may result in challenging daytime behaviors [23]. There is a correlation between sleep problems and increased aggression, noncompliance, increased social skills deficits, an increase in emotional behaviors, deficits in daily life skills [41], and a decrease in communication skills [19]. Insufficient sleep has a profound negative effect on daily activities and social inclusion for those affected. Disrupted sleep patterns experienced by children with ASD/ID and the subsequent negative impact on their families can result in a reduced quality of life for the family unit. Furthermore, sleep problems in children with or without ASD/ID are associated with increased parental stress, reduced parental sense of competence, and poor physical health for the family unit [32].

Individuals with ASD/ID require effective user-friendly sleep interventions that meet their diverse needs. “User-friendly” within the current context refers to well-designed individualized behavioral strategies that parents or carers/caregivers find easy to implement within the home and family routines. Sleep interventions can be broadly categorized into three approaches: namely, pharmacological, behavioral, or combined pharmacological and behavioral. Pharmacological interventions aim to increase sleep duration and to decrease sleep latency. However, sleep-promoting medications may have numerous side effects and could have an adverse interaction with other medications [12]. It is suggested that melatonin may be an effective pharmacological approach for children with neurodevelopmental disorders with minimal adverse effects [5]. Melatonin treatment may benefit children with developmental disorders such as ASD/ID to decrease sleep onset latency and to increase total sleep time; nonetheless, it is argued that it will not decrease nighttime awakenings [9]. As the current review was not investigating the efficacy of pharmacological sleep treatments, those studies that were purely pharmacological were excluded.

Research has suggested that behavioral interventions are effective for improving a range of sleep problems experienced by individuals with developmental disorders [18,23,39]. Behavioral interventions can include a range of procedures to include sleep hygiene, reinforcement of desirable behavior, ignoring undesirable behaviors, and environmental changes. They encourage the initiation and maintenance of sleep and are recommended as the first approach to resolve sleep disturbances in children [29]. Behavioral interventions aim to increase the frequency of behaviors that are crucial to improving sleep and to decrease the behaviors that are detrimental to sleep. Many sleep interventions for children with or without ASD/ID are implemented within the home environment by parents. Sleep interventions can be time consuming and involve ignoring unwanted bedtime behaviors which the parent may find stressful, such as the child crying out. It is important that parents are given the knowledge, support, and appropriate training on how to deal with undesirable bedtime behaviors (eg, how to change the environment and how to deliver timely reinforcement to enhance the effectiveness of the behavioral sleep intervention). Behavioral interventions incorporating parent training are increasing, and in recent years parent training has been introduced to enhance the treatment of sleep problems for children with ASD/ID [6,32,42,45].

Several reviews have been published relating to insomnia in children with ASD/ID and atypically developing children. However, only three of the most recent reviews included a few studies that were relevant to the current systematic review inclusion criteria of parent training. Vriend et al., [47] published a review on the effectiveness of 15 behavioral interventions for sleep problems in children with ASD with studies dating from 1964 to 2010. Five of the studies included by Vriend et al., published between 2004 and 2010, incorporated an element of parent training or sleep education. One study [31] was of interest to the current review, wherein the researchers conducted sleep interventions using parent

training and behavioral strategies with three children with ASD. It could be argued that knowledge of sleep education, behavioural strategies and guidance on how to implement them within the home environment was invaluable to achieving the positive outcomes reported by Moon et al., [31]. Vriend et al., concluded that there was evidence to suggest that some behavioral interventions focusing on sleep problems were possibly effective for children with ASD/ID. This conclusion led them to advocate the need for further research to examine the effectiveness of evidence-based sleep interventions for children with ASD/ID, using larger samples within rigorously controlled designs.

Malow et al., [24] published a review on the identification, evaluation, and management of insomnia in children and adolescents with ASD. They included behavioral, alternative, and pharmacological publications dated from 2000 to 2011. Only one of these studies [37] was relevant to the current review. Reed et al., [37] had a within-subject design that implemented a multi-component behavioral parent training programme to improve a wide range of sleep problems experienced by children with ASD. They [37] evaluated the use of three face-to-face group workshops with three to five families per session, in which they used a standardized training programme with opportunities for individualization. Sleep improvements were assessed using questionnaires and actigraphy. Results demonstrated a reduction in the target behaviors of bedtime resistance, initiating sleep, night waking, or early waking in many of the children. These findings may suggest that behavioral parent training, if utilized and maintained, could be a suitable and effective addition to enhance parents' confidence and ability to apply behavioral sleep intervention procedures. Malows et al., concluded that parent sleep education training combined with behavioral techniques should be the first approach to improving sleep difficulties in children with ASD.

A third review, conducted by Meltzer and Mindell [28], on behavioral interventions for paediatric insomnia focused mainly on typically developing children and included publications from 1985 to 2013. Although their review included two studies with a special needs population, only one of these studies [1] was applicable for inclusion within the current systematic review. In conclusion, Meltzer and Mindell highlighted the need for further evidence on the efficacy of behavioral interventions for pediatric insomnia.

Considering the evidence presented in the three previously summarized reviews, the time frame for the current systematic review was set between 2010 and 2016. Comprehensive electronic database searches for the six-year time frame for the current review identified 11 new publications that included parent training. Notably, the number of studies published had increased since the Vriend et al., [47] review, in which, over a similar time frame of six years from 2004 to 2010, five publications were cited that included an element of parent training. This could possibly signify a change in the trend towards an increase in behavioral sleep interventions that include parent training.

1.1. Rationale and objectives for current review

There is a need for the identification and synthesis of outcomes from evidence-based behavioral practices using parent training to improve sleep in children with ASD/ID. It is equally important to evaluate the quality and mode of delivery and to reflect on the variations in the methods employed within these interventions. In doing so, knowledge, impact, and societal value are all enhanced, enabling clinicians and families to make informed choices. Further to this, the current review may inform and guide future research directions and clinical practice in the management of pediatric sleep problems. This article presents the first systematic review to focus on parent training incorporated

within behavioral sleep interventions for children with ASD/ID. In addition, it updates the existing review literature by including studies published between 2010 and 2016. Thus, the goal of this article was to identify and to systematically review the evidence on the efficacy of current behavioral interventions that include parent training in the treatment of sleep problems experienced by children with ASD/ID.

2. Method

2.1. Eligibility criteria

Upon receipt of ethical approval from the NUI Galway Research Ethics Committee, the following inclusion criteria were implemented: (1) included a sample that was representative of children with ASD and/or ID who experienced sleep problems; (2) combined behavioral strategies with parent training, parent-mediated intervention, or parent sleep education in the treatment of sleep problems; (3) research designs included single-group design (SGD), randomized and nonrandomized controlled trials (RCTs and non-RCTs, respectively), single-subject research designs (SSRDs), and no experimental case studies using pre and post measurements; (4) published between January 2010 and November 2016; (5) published in peer-reviewed journals; and (6) published in the English language. Exclusion criteria were as follows (1) purely pharmacological sleep treatments; (2) studies that did not include parent sleep education, training, or parental mediated interventions; (3) if the sample was not identified as ASD/ID; (4) if the sleep intervention was implemented with an adult population; and (5) all reports, reviews, dissertations, and poster presentations were excluded.

2.2. Search procedures

During 2016, the three electronic databases of PsycINFO, PubMed, and Web of Science were searched to identify potential studies. Keyword search terms included three areas related to autism (autis* OR pervasive develop* OR Asperger* AND learning disabilities OR intellectual disability*), sleep disorders (sleep problems* OR sleep disorders OR sleep difficulties), and interventions (behavioral intervention AND training OR parent* OR education OR mediated). The database searches resulted in 2765 potential publications. Five additional studies were identified by manually scanning the reference lists of the full-text publications which met the inclusion criteria for the current review.

2.3. Study selection

After exporting all publications to a reference management software, 602 duplicated articles were removed and the remaining 2163 titles and abstracts were screened for eligibility. This resulted in the removal of a further 1749 studies based on the exclusion criteria listed earlier. The remaining 416 full-text publications were assessed for eligibility, and 11 studies were identified as meeting the inclusion criteria listed earlier (see Fig. 1 for a flow diagram summarizing this process). The first and second authors independently screened the 11 studies using the inclusion and exclusion criteria for this review, and there was 100% agreement for the inclusion of all 11 studies.

2.4. Methodological quality

The first and second authors independently assessed the methodological quality of the 11 included studies using the Quality

Index [13]. The Downs and Black Quality Index [13] has been shown to provide a profile of randomised and non-randomized studies. It has been extensively used by reviewers to highlight the methodological strengths and weaknesses of health care studies. The scale consists of five subscales to include reporting (10 items), external validity (three items), bias (seven items), confounding (six items), and power (one item). The items are answered as 'yes', 'no', 'partially', or 'unable to determine' depending on the subscale. Responses are scored 0, 0, or 1, except for question 5 (reporting subscale) which can be scored 0, 1, or 2, and question 27 (power subscale), which can be scored 0, 1, 2, 3, 4, or 5. A maximum overall score of 32 can be achieved, with higher scores indicating higher quality. The index can also be scored and summed to produce five subscale scores. Internal consistency for the Quality Index is high (KR-20 = 0.89) with RCTs (KR-20 = 0.92) and non-RCTs (KR-20 = 0.88). Test-retest reliability ($r = 0.88$, $p = 0.90$), with RCTs ($r = 0.76$, $p = 1.00$) and non-RCTs ($r = 0.7$, $p = 0.84$) and interrater reliability ($r = 0.75$, $p = 0.056$) of the Quality Index are good [13]. Interrater reliability (IRR) for the current review was calculated for each subscale (reporting 93% IRR, external validity 96% IRR, internal validity bias 91% IRR, confounding and selection bias 97% IRR, and power 100% IRR), and the resulting overall quality score (96% IRR) for each study (see Table 1 for reviewers' independent scores). Although the reviewers' scores varied slightly on occasion, their overall quality assessment score agreed as to which methodological quality category matched the study (100% IRR). The results indicated that nine studies were of high quality, three studies were of good quality, and that no studies were rated as of adequate or poor quality.

2.5. Data extraction

Data extraction for all included studies was completed by the first and second authors using an adapted version of the best-practice data collection form for reviews incorporating a range of methodological designs (downloaded from <http://epoc.cochrane.org/resources>) [14]. Data extraction included the number of participants, age range, gender, sample population, that is, ASD and/or ID, diagnosis criterion, setting, recruitment method, and type of sleep problem reported. Along with study design (SGD, RCTs, non-RCTs, SSRDs, or case studies using pre and post measurements), type of parent involvement (parent training, sleep education, or parent-mediated intervention) and mode of training delivery (telephone, face-to face, group, individual, workshop, or leaflets) were noted. Data for attrition rates, treatment integrity, measurements employed, and intervention outcomes were also extracted. Interrater reliability for data extraction was high, at 94%. A summary of the extracted data are presented in Table 2.

3. Results

3.1. Narrative synthesis of participants and study design

Across the 11 studies, a total of 416 children and 414 parents participated; in the eight studies that specified the child's gender, 188 of 250 children (75%) were male. Stuttard et al., [42] included young children aged 1–4 years with a mean age of three years. Most of the studies provided data on children with ages ranging from 2 to 11 years with a mean age of five years, with exception of [32] and [42], which included children and adolescents aged 8–17 years with a mean age of 12 years, and 5–15 years with a mean age of nine years, respectively. Ten of the studies included children ASD/ID, whereas Allen et al [2] study was specific to Angelman syndrome. Four of the 11 studies also included children with ID, Down syndrome, cerebral palsy, learning disability, and decreased

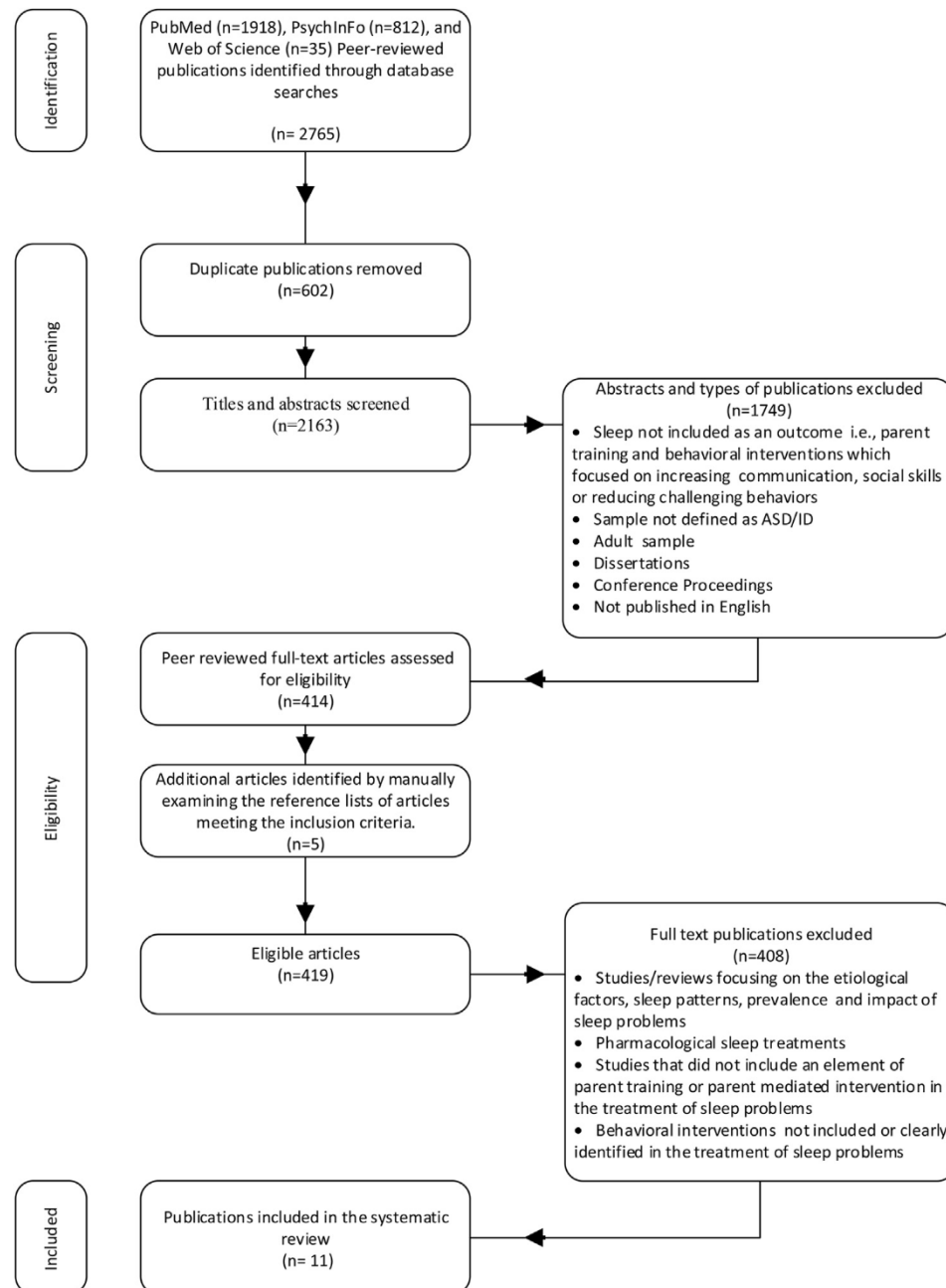


Fig. 1. Flow diagram depicting the identification and selection process for included review studies.

intellectual functioning with an IQ score of ≤ 70 . A professional assessment and diagnosis were provided for all children included in the current review studies.

Of the 11 studies, four (36.3%) were RTCs, of which two included waitlist control groups and two involved different treatment groups. Of the remaining studies, three (27.3%) were single-group design, two (18.2%) were comparison-group designs, one (9.1%) was a single-subject design, and one (9.1%) was a case series design (Table 2). Methodological quality assessment indicated that nine studies were of high quality and three studies were of good quality, as summarized in Table 1.

Studies included in this review differed considerably in their experimental design, sample population, study duration, parent contact time with professional support, parent training programs,

and training mode of delivery. Nonetheless, nine of the 11 studies had similar aims to reduce sleep difficulties experienced by children with ASD/ID by means of behavioral sleep interventions implemented by parents who were supported with parent training and/or sleep education materials.

3.2. Parent training programs and sleep outcomes

In Adkins et al., [1], parents of children ($n = 36$) with ASD, Asperger's and Pervasive Development Disorder-Not Otherwise Specified (PDD-NOS) who experienced sleep onset latency of 30 min or more were randomly stratified by their child's age into the treatment group ($n = 18$) and wait list control group ($n = 18$). A sleep education pamphlet developed by the Autism Treatment

Table 1

First and second authors' independent scores for the Downs & Black Quality Assessment Scale for included studies.

Authors	Reporting	Reporting	External validity	External validity	Internal validity	Internal validity	Confounding and selection bias	Confounding and selection bias	Power	Power	Total Quality Rating Score [13]	Total Quality Rating Score [13]
	First	Second	First	Second	First	Second	First	Second	First	Second	First	Second
Moon et al. [31]	10/11	11/11	3/3	3/3	4/7	4/7	4/6	4/6	1/5	1/5	22/32 Good	23/32 Good
Adkins et al. [1]	10/11	10/11	3/3	3/3	4/7	4/7	3/6	3/6	5/5	5/5	25/32 High	25/32 High
Allen et al. [2]	9/11	9/11	1/3	1/3	3/7	2/7	4/6	3/6	2/5	2/5	19/32 Good	17/32 Good
Austin et al. [6]	10/11	11/11	3/3	2/3	3/7	3/7	2/6	2/6	5/5	4/5	23/32 Good	22/32 Good
Johnson et al. [21]	11/11	11/11	3/3	3/3	6/7	7/7	6/6	6/6	5/5	5/5	31/32 High	32/32 High
Malow et al. [23]	11/11	11/11	3/3	3/3	6/7	7/7	6/6	6/6	5/5	5/5	31/32 High	32/32 High
Moss et al. [32]	10/11	11/11	3/3	3/3	6/7	6/7	6/6	6/6	5/5	5/5	30/32 High	31/32 High
Fawkes et al. [15]	10/11	11/11	3/3	3/3	6/7	7/7	5/6	6/6	5/5	5/5	29/32 High	32/32 High
Stuttard, Beresford et al. [42]	10/11	11/11	3/3	3/3	4/7	5/7	3/6	3/6	5/5	5/5	25/32 High	27/32 High
Stuttard, Clarke et al. [42]	10/11	11/11	3/3	3/3	5/7	5/7	4/6	4/6	5/5	5/5	27/32 High	28/32 High
Veatch et al. [45]	10/11	11/11	3/3	3/3	5/7	7/7	5/6	6/6	5/5	5/5	28/32 High	32/32 High

First, first author; second, second author.

Network Sleep Committee was distributed to parents in the treatment group. Treatment group parents were instructed to read the pamphlet. No further support was given to the parents during the study. It was reported that the use of the pamphlet did not conclusively improve sleep within this sample. Feedback from parents suggested that the pamphlet was informative but lacked guidance on how to put the information into practice.

Two studies Moss et al., [32] and Austin et al., [6] investigated the 'Sleepwise' program, with children ($n = 26$) with ASD, Angelman Syndrome, Down Syndrome and ID [32] and with children ($n = 8$) with ASD or PDD-NOS [6]. Moss et al., [32] used an RTC design to compare a parent training workshop ($n = 13$) with a wait list control group ($n = 13$), and reported improvements in sleep problems for both groups, although the improvements were significantly greater for the 'Sleepwise' treatment group than for the wait list control group. In contrast, Austin et al., used a single-group design to evaluate the same program and reported significant sleep improvements [6].

Managing Your Child's Behavior to Promote Better Sleep (MCBPBS) training manual [42], and the Parent Education Curriculum Training program [23,45] were associated with positive outcomes. All three studies reported significant success in improving sleep outcomes for their sample group. Stuttard et al., [42] further observed that parents' attitudes and confidence had improved after training. In addition, Veatch et al., [45], reported that the parent sleep education training resulted in closer correlations between the subjective and objective sleep measures employed during the study.

Allen et al., [2] found that a Behavioral Treatment Package (BTP) was effective for all children ($n = 5$) with Angelman syndrome who experienced prolonged sleep difficulties. Each child learned to initiate falling asleep independently, and the duration of sleep was increased on average by 30 min per night. Parents also reported that there was a marked improvement in their child's disruptive bedtime behaviors. Allen et al., concluded that enabling the parents

to enhance the sleep environment, modify sleep schedules, and alter parent–child interactions during bedtime routines and throughout the night resulted in immediate improvements in the child's sleep patterns.

Moon et al., [31] evaluated the effectiveness of the 'Better Nights, Better Days' parent handbook to reduce sleep onset latency in children ($n = 3$) with ASD. The mean sleep onset latency decreased for all three children; although the improvements were relatively small, they were maintained at the follow-up assessment. However, for two of the three children, the average duration of sleep decreased slightly.

3.3. Training delivery mode and professional support

Within the current sample of studies, the parent training delivery mode included group ($n = 4$), individual ($n = 5$), or self-directed ($n = 2$). Ten of the studies offered varying degrees of professional support/contact throughout the intervention; however, one study, by Adkins et al., offered no further support to parents after the distribution of the sleep education pamphlet [1]. In Adkins et al., the self-directed support approach revealed that parents found the pamphlet informative but lacked guidance on how to put the information into practice.

Fawkes et al., [15] compared the data from two previously published studies that used different protocols. The first study used brief verbal training during an informed consent visit for children ($n = 20$) with ASD experiencing sleep problems [37]. The second study delivered a 1-h structured hands-on training prior to commencement of the sleep intervention for children ($n = 80$) with ASD, Asperger's disorder, and PDD-NOS [23]. Fawkes et al., focused primarily on improving the scoring on the actigraphy and sleep diary measurements, which are essential elements for ascertaining the efficacy of sleep interventions [15]. Although Fawkes et al., did not measure sleep outcomes directly, the study did produce

Table 2

Summary of research studies included in the current systematic review.

Author/s	Participants' age/gender/diagnosis	Design	Sleep issues	Measurement	Intervention	Results	Quality Rating [13]
Moon et al. [31]	Parents (n = 3) & children (n = 3: 8–9 years: 2 males & 1 female). Autism Spectrum Disorder.	Case series design.	Sleep onset latency, sleep duration & sleep efficiency.	Pre, Post & Follow up. Actigraphy, Sleep diaries, CSHQ, CBC & Parent Satisfaction Questionnaire.	Five weeks' self-delivered: Better Nights, Better Days: Treatment for sleep difficulties parent handbook. Telephone support. Behavioral strategies: Faded bedtime, Response Cost & Positive reinforcement.	Mean sleep onset latency was reduced for all 3 children. Maintained at 12 weeks follow up. Parents satisfied. Parent handbook considered to be effective.	Good
Adkins et al. [1]	Parents (n = 36) & children (n = 36: 2–10 years: 24 males and 12 females). Autism Spectrum Disorder, Asperger's & Pervasive Development Disorder -Not Otherwise Specified.	RCT Sleep Education Pamphlet (n = 18) Control Group No Pamphlet (n = 18).	Sleep onset latency, sleep duration, sleep efficiency & wake time after sleep onset.	Pre, Post & Follow up. Actigraphy, CSHQ & Sleep diaries, Stanford-Binet 5, Mullen Scales of Early Learning.	Distribution of Sleep education pamphlet developed by Autism Treatment Network Sleep Committee. Parents received no further support throughout the duration of the study.	Use of the pamphlet did not conclusively improve sleep within this sample. Feedback from parents indicated information was valuable but guidance on how to implement the strategies would be beneficial.	High
Allen et al. [2]	Parents (n = 5) & children (n = 5: 2–11 years; 3 females & 2 males). Angelman syndrome.	Multiple Baseline SSRD.	Sleep onset latency, sleep duration, sleep efficiency & wake time after sleep onset. Initiate sleep independently.	Pre, Post & Follow up. Actigraphy, CSHQ & Sleep diaries, Developmental Behavior Checklist, Abbreviated Acceptability Rating Profile.	Parent mediated Behavioral Treatment Package (BTP). To include sleep environment, sleep-wake schedule & parent/child interactions. Professional support throughout via telephone or video contact.	The BTP was found to be effective for all 5 children who had experienced prolonged sleep problems.	Good
Austin et al. [6]	Parents (n = 6: 2 males & 4 females) & children (n = 8: 3–7 years: 8 males). Autism Spectrum Disorder, Pervasive Development Disorder & PDD-Not Otherwise Specified.	Single group (n = 6) for parent training & SSRD for sleep interventions.	Night waking, Bedtime resistance, co-sleeping, early waking & irregular sleep-wake cycle.	Pre, Post & Follow up. CSHQ, DBC-P, Sleep diaries, Sleepwise Sleep Disturbance Index, Caregivers Acceptance of Treatment Survey (CATS).	A 15-week Sleepwise program consisted of 3 x 2-h workshops focusing on sleep education and behavioral strategies. Along with ongoing support during sleep interventions.	Sleepwise program was effective with all 8 children making significant improvements in sleep problems.	Good
Johnson et al. [21]	Parents (n = 40) & children (n = 40: 2–6 years). Gender not reported. Autism Spectrum Disorder, Pervasive Development Disorder -Not Otherwise Specified.	RCT Behavior Parent Training (n = 20) Psycho-educational training (n = 20).	Sleep onset delay, bedtime resistance, night awakening & early morning awakening.	Pre, 4 weeks & 8 weeks. Actigraphy, Composite Sleep Index, Sleep diaries, Treatment fidelity checklist, Parent satisfaction.	Five individual sessions (60–90 min) over 8 weeks were delivered by 2 Board Certified Behavior Analysts and the third therapist was training with the BACB. The BPT included specific bedtime and sleep management training whereas the PE training related to ASD diagnosis and developmental issues.	Mixed results with the actigraphy not detecting a significant difference between the two groups. Although the BPT group indicated via the Composite Sleep Index that there was a significant improvement in bedtime and sleep behaviors.	Good

Table 2 (continued)

Author/s	Participants' age/ gender/diagnosis	Design	Sleep issues	Measurement	Intervention	Results	Quality Rating [13]
Malow et al. [23]	Parents (n = 80) & children (n = 80: 2–10 years: 64 males & 16 females). Autism Spectrum Disorder. Asperger's, PDD-NOS & Intellectual Disability.	RCT Individual Education Program (n = 40) & Group Education Program (n = 40).	Sleep onset delay, Sleep efficiency & wake time after sleep onset.	Pre & Post. Actigraphy, Sleep diaries, CSHQ, FISH, CBCL, RBS-R, Peds QL, PSOC & End of Education Session Survey.	Group (n = 2 to 4 parents: 2 x 2-h sessions over 2 weeks) & Individual one x 1-h session. Parent education curriculum training program [37]. Telephone follow-up. Behavioral strategies included graduated extinction paired with rewards, bedtime pass, visual schedules & sleep hygiene.	Improvements observed in sleep problems and behavioral measures regardless of the mode of parent education.	High
Moss et al. [32]	Parents (n = 26: 29–61 years) & children (n = 26: 8–17 years). Gender not reported. Autism Spectrum Disorder, Angelman Syndrome, Down Syndrome, Intellectual Disability & Blindness.	RCT <i>Sleepwise</i> treatment Group (n = 13) & Wait List Control Group (n = 13).	Night waking, sleep duration, settling to sleep, excessive daytime sleepiness, co-sleeping and early waking.	Pre, Post & Follow-up. CSHQ, DBC-P, PSI-SF, Consumer Satisfaction using a semi structured interview, CATS & Goal Attainment Scale.	A 10-week <i>Sleepwise</i> program (adapted for older children & teenagers) consisted of 3 x 2-h workshops (n = 5–7 parents) focusing on sleep education and behavioral strategies. Professional facilitators supported the program throughout within the workshops; home observations and visits.	<i>Sleepwise</i> program was effective. Both groups reduced sleep problems however the improvements were significantly greater for the treatment group compared to the WL.	High
Fawkes et al. [15]	Study 1: Pilot study Parents (n = 20) & children (n = 20: 4–10 years) Gender not reported. Autism Spectrum Disorder [37]. Study 2: Parents (n = 80) & children (n = 80: 2–10 years). Gender not reported. Autism Spectrum Disorder, Asperger's & Pervasive Development Disorder -Not Otherwise Specified [23].	Comparison group design between study 1 (n = 20) conducted by Ref. [37] & study 2 (n = 80) Conducted by Ref. [23].	Specific sleep problems were not reported in Ref. [15].	Pre & Post Actigraphy Sleep diaries	Comparing two methods of parent training on the use of the actigraphy and sleep diaries. Brief verbal training during study one's informed consent visit versus 1-h structured hands on actigraphy and sleep diary training visit before study two commenced.	A higher number of nights were scored on the actigraphy and sleep diary data for study two. This was attributed to the structured educational hands on parent training delivered during study two.	High
Stuttard, Beresford et al. [42]	Parents (n = 23) & children (n = 22: 5–15 years: 13 males & 9 females). Autism Spectrum Disorder; Intellectual Disability	Single group design (n = 23).	Bedtime resistance; night awakening and night-time self-settling.	Pre, Post & Follow up at 3 & 6 months. CSHQ, PSCO, Implementation fidelity checklist.	Group delivered (n = 4–9 parents) in one of the 4 x 3 h sessions over 5 weeks. Two learning disability nurses form CAMHS-LD team delivered the Managing your child's behavior to promote better sleep (MCBPBS) manual.	Improvements in child's bedtime resistance and night awakenings were observed. Parent's beliefs, attitudes and confidence improved.	High

(continued on next page)

Table 2 (continued)

Author/s	Participants' age/ gender/diagnosis	Design	Sleep issues	Measurement	Intervention	Results	Quality Rating [13]
Stuttard, Clarke et al. [42]	Parents (n = 15) & children (n = 15: 1–4 years: 11 males and 4 females). Autism Spectrum Disorder, Cerebral Palsy, Developmental Delay, Sensory Impairments & Learning Disability.	Qualitative analysis. Comparison groups: home visits (HV n = 7) & telephone contact (TC n = 8) & SSRD for sleep interventions.	Sleep problems.	Pre, Post & Follow up at 2–3 months. Semi structured interviews.	Specialist health visitors provided Sleep strategy implementation support (SSIS) to deliver an Intensive behavioral sleep management intervention (IBSMI). Individual HV (3–9 weeks for 30 min to 1 h) or TC (3–10 weeks for 10 and 30 min).	Irrespective of the delivery mode (HV or TC) parents reported benefits from receiving SSIS to implement the IBSMI. TC was viewed to be an acceptable and convenient mode to deliver SSIS.	High
Veatch et al. [45]	Parents (n = 80) & children (n = 80: 2–10 years: 64 males & 16 females) Autism Spectrum Disorder.	Single group design.	Sleep duration, bedtime resistance, night time awakenings.	Pre & Post. Actigraphy, CSHQ, sleep dairies.	Parent education curriculum training program (Malow et al., [23]). Good habits that contribute to good sleep patterns, appropriate timing of sleep, visual schedules, healthy child/parent interactions during night time awakenings.	Parent sleep education resulted in improvements in correlations between objective and subjective measurements of sleep problems.	High

CBC, Child Behavior Checklist; CSHQ, Child Sleep Habit Questionnaire; RCT, randomized control trial; SSRD, single-subject research design.

Note. RCT – Randomized Control Trials, CSHQ – Child Sleep Habit Questionnaire, DBC-P – Developmental Behavior Checklist-Parent version, RBS-R – Repetitive Behavior Scale-Revised, FISH – Family Inventory of Sleep Habits, CBC – Child Behavior Checklist, PedsQL – The Parent Proxy-Report of the Pediatric Quality of Life Inventory, PSOC – Parents Sense of Competence Scale, PSI-SF – The Parenting Stress Index: Short Form.

Note. RCT – Randomized Control Trials.

Note. SSRD – Single Subject Research Design, CSHQ – Child Sleep Habit Questionnaire, PSOC – Parents Sense of Competence Scale, CAMHS-LD – Child and Adolescents Mental Health Service – Learning Disability.

evidence to support the suggestion that hands-on extended parent training was more valuable than the brief verbal instructions [15].

Stuttard, Clarke, Thomas and Beresford, [42] conducted a study with children (n = 15) with ASD, Cerebral Palsy, Developmental Delay, Sensory Impairments and Learning disability which implemented an individualized intensive behavioral sleep management intervention. Parents received continued support either through home visits (n = 7) or telephone calls (n = 8). Even though Stuttard et al., did not measure sleep as an outcome, they did provide some evidence of the effectiveness of the individual delivery mode irrespective of the type of professional contact [42]. It was found that training was beneficial and valued by the parents implementing sleep interventions.

Two further studies evaluating the mode of delivery of parent training programs were Malow et al., [23] for children (n = 80) with ASD, Asperger's, PDD-NOS, and ID, and Johnson et al., [21] for children (n = 40) with ASD and PDD-NOS. Malow et al., [23] compared individual parent education training (n = 40) with group parent education training (n = 40), whereas Johnson et al., [21] compared individual delivery mode of behavior parent training (BPT, n = 20) with individual psycho-educational parent training (PE, n = 20). Malow et al., [23] observed improvements in sleep outcomes regardless of the delivery mode of parent education training. In contrast, Johnson et al., [21] reported mixed results. Measurements obtained using the actigraphy indicated no differences between the BPT and PE groups, although measurements provided by the Composite Sleep Index indicated that there was a significant improvement in bedtime and sleep behaviors for the BPT group in comparison to the PE group. In terms of professional support, participating parents in the Johnson et al., study attended five individually administered 60- to 90-min sessions with a

trained sleep therapist in a clinic setting [21]. In comparison, parents participating in the Malow et al., [23] study were supported by trained sleep educators, with the group education program providing two 2-h sessions over two weeks compared to a single 1-h session provided by the individualized program. All parents received two follow-up phone calls.

Moss et al., [32] and Austin et al., [6] (as detailed in parent training programs and sleep outcomes above), offered parent training using a group mode of delivery which consisted of three 2-h sessions. However, the duration of the training differed, with Austin et al., [6] running the program for 15 weeks as opposed to Moss et al., [32] running it for 10 weeks. Professional facilitators supported both programs throughout, within the workshops and with home observations and visits. Both studies reported positive outcomes from the parent training program.

3.4. Follow-up and maintenance

Five of the 11 studies reported follow-up data on sleep outcomes; these follow-up periods ranged from one month to six months postintervention. All five studies reported that improvements in sleep outcomes were maintained at a one-month follow-up [6], after two months [32], from one to three months [2], after three months [31], and for repeated follow-ups at three and six months [42].

3.5. Treatment fidelity and social validity

Several of the studies included in the current review, with the exception of Fawkes et al. [15] and Veatch et al. [46], reported either treatment fidelity and/or social validity data. Stuttard, Beresford et

al., [42] reported implementation fidelity data for the content of the training workshops which were rated 100% consistent, relevant, beneficial and effective. Parent attendance was generally good, with 11 parents attending all four workshops, and the acceptability of the training workshops was considered promising.

In a study by Johnson et al., [21], treatment fidelity measured the delivery of treatment as intended and parent adherence to delivered treatment across the two treatment groups of BPT and PE. Results were high for both groups, with BPT at 98% for treatment as intended and 93% for parent adherence to delivered treatment; the PE was 99% for treatment as intended and 98% for parent adherence to delivered treatment. Interrater reliability for treatment as intended was reported as 98% for BPT group and 99% for the PE group, and parent adherence to the delivered treatment was 91% for the BPT group and 98% for the PE group. A total of 30 parents provided social validity data by completing a parent's satisfaction questionnaire, on which 90% satisfaction was reported for the BPT group and 88% for the PE group.

Of the remaining seven studies that provided social validity data [2], found that parents evaluated the BTP as highly acceptable and effective, and stated that they intended to continue to use the BTP. The parents who participated in the [32] study were satisfied with the intervention outcomes and indicated that the Sleepwise program was acceptable, valuable, and effective. The Sleepwise program was also evaluated in the [6] study, and parents found that the resources used, training delivered, and outcomes were beneficial and effective. In addition, parents stated that peer support was an influential factor throughout the Sleepwise program. Parent satisfaction measured in the Moon et al., study indicated that parents were satisfied with the sleep intervention and that the manual was helpful [31]. Adkins et al., [1] received feedback from parents that suggested that the pamphlet was informative but lacked guidance on how to put the information into practice. In the Malow et al., [23] the parent's evaluation of the overall programme and the educators' training delivery mode indicated a high level of satisfaction with 75% for the group delivery sessions and 86% for the individual sessions. In total, 91% of parents agreed that there were improvements in sleep habits. Parents' reported views in the Stuttard, Clarke et al., [42] revealed that parents felt the telephone contact was considered acceptable and less time consuming nonetheless the telephone calls were considered very impersonal. Parents also expressed that the initial home visit to instruct the TC group on how to implement the sleep intervention strategies was essential to the success of the intervention. It was expressed that more face-to-face contact would have been valuable.

4. Discussion

The aim of the present review was to evaluate and to synthesize the effectiveness of parent training programs incorporated in behavioral sleep interventions for children with ASD and/or ID. In the limited research currently available, it is evident that parent training programs have the potential to reduce prevalent sleep problems such as initiating and maintaining sleep, early morning and night awakenings, and bedtime resistance. Future research should build on this evidence to first clarify which elements of parent training improve acquisition of healthy sleep practice knowledge, and second, to identify the challenges that may exist which prevent parents from implementing sleep knowledge into healthy sleep practice.

4.1. Current evidence

Eight of the nine studies that measured sleep behaviors as an outcome repeatedly demonstrated a reduction in the target

behaviors for several of the children. There are numerous factors that may have contributed to the meaningful sleep outcomes reported in the studies included in this review. Parent training programs included across the 11 studies incorporated a multi-element approach with a combination of the following components of sleep hygiene practice, sleep education, reinforcement, extinction, faded bedtime, response cost, sleep–wake schedule, parent–child nighttime interaction, and psychoeducation. It is possible that the improvements were assisted by the parent training program content, the duration and mode of delivery of the parent training, along with professional support received, for example, face-to-face consultation versus written information or group versus individual contact.

Notably, four of the studies included in the current review [1,6,31,32] used written parent training materials. The content of the Sleepwise manual employed by Moss et al., and Austin et al., as well as the Better Nights, Better Days manual used by Moon et al., [31] included sleep education and behavioral strategy topics similar to those presented in the standardized pamphlet for insomnia in children with ASD used by Adkins et al., [1]. However, Adkins et al. reported negative results and Moss et al., Austin et al., and Moon et al. reported positive results. Adkins et al. found the pamphlet to be ineffective for insomnia in children with ASD, whereas Moss et al., [32] Austin et al., [6] and Moon et al., [31] reported significant positive sleep outcomes for children with developmental disabilities and ASD, respectively. Albeit not conclusive, it is possible that the different delivery modes implemented, and the level of professional support received, could have contributed to the reported outcomes. Moss et al., [32] and Austin et al., [6] provided professional facilitators who supported the program with home visits and observations, and all parents attended three 2-h workshops which focused on sleep education and behavioral strategies. Moon et al., [31] supported the parents via weekly telephone calls for the duration of the study. In contrast, Adkins et al., [1] distributed the sleep education pamphlet and the parents received no further support for the duration of the study. The difference in the delivery mode and/or the insufficient professional contact/support may be an explanation for the negative results reported by Adkins et al., [1]. These results would suggest that factors such as the delivery mode, advice, and ongoing support are essential to maximize treatment outcomes.

Further evidence presented from studies evaluated within the current review would suggest that training delivered either in a group or in individual format produced similar positive treatment outcomes [23]. Three studies also produced positive outcomes by delivering parent training sessions ranging from two to five weeks with durations varying from 1 to 4 h [21,23,42]. In addition, parent training that was delivered by professionals and that contained similar sleep education and behavioral intervention content produced positive treatment outcomes [21,23,42]. The synthesis of these studies has provided useful information on the program content, mode of delivery, and professional sleep educators' input into the implementation of evidence-based parent training that can improve sleep interventions for children with ASD/ID.

In addition, two comparison group studies conducted by Fawkes et al., [15] and Stuttard, Clarke, et al., [43] did not report evidence for sleep outcomes. However, these authors did provide additional evidence to support the efficacy of parent training for sleep problems experienced by children with ASD/ID. This evidence offered information regarding the level of professional support options which could be successfully integrated into behavioral sleep interventions. Fawkes et al., produced evidence from two previously published research articles to support the suggestion that hands-on extended parent training was more valuable than brief verbal instructions [15]. As for Stuttard, et al., [14] they found that parent

training was beneficial irrespective of the type of support offered during training and home implementation stage, for instance, home visits (HV) or telephone calls (TC). The defining difference between the two studies was that Stuttard, et al., [14] provided all parents with an intensive behavioral sleep management intervention, with the HV group receiving 30- to 60-min sessions for 3–9 weeks and the TC group receiving 10–30 min for 3–10 weeks before delivering the home sleep strategy implementation support. In Fawkes et al., [15] data from previous research were compared wherein one group of parents were offered face-to-face extended training and the other group of parents were offered brief verbal instructions. It would appear from the evidence presented in these two studies that face-to-face, good-quality training surpasses brief verbal instructions and that parents did not differentiate between HV and TC and viewed both as acceptable methods of support. It is always difficult to rule out alternative explanations for conflicting evidence, and this is one area of research that could benefit from future investigation.

4.2. Quality of evidence

Methodological quality was assessed using the Downs and Black [13] assessment criteria, which revealed that eight of the included studies were of high quality and three were of good quality. The benefits of using the Downs and Black assessment was that it provided a more objective approach to the quality of the evidence presented within the current review. Rather than solely relying on each of studies' reported successful outcomes, the strength of the evidence was also based on the quality of report writing, external validity, internal validity, confounding and selection bias, and statistical power for each study. Studies with high methodological quality will enhance the evidence and thus help to establish and promote the approach as evidence-based practice for effective behavioral sleep interventions for children with ASD/ID.

4.3. Follow-up and maintenance

Not all the studies included in the current review reported follow-up and maintenance data on treatment outcomes. Within the current review, five of the 11 studies provided short-term to relatively long-term data on sleep outcomes. Each of the five studies reported that improvements in sleep outcomes were maintained at a one-month follow-up [6], after two months [32], from one to three months [2], after three months [31], and for repeated follow-ups at three and six months [43]. The number of studies that did not report follow-up efficacy on treatment outcomes was a concern highlighted by the current review. To consider an intervention as effective, it is imperative that the posttreatment long-term positive effects are maintained. The current review was unable to conclusively offer a strong synthesis of the maintenance data. From an effective treatment perspective, future research should consider incorporating data collection on treatment outcome maintenance into research procedures. Considering the evidence that parenting practices contribute to the improvement of sleep problems [2,6,21,23,31,32,42,43]. An additional topic of interest should be the follow-up assessment of the maintenance of parental knowledge/practice, as this can have implications for either retaining sleep improvements or further enhancements in sleep outcomes.

4.4. Treatment fidelity and social validity

Several of the studies included in the current review, with exception of Fawkes et al., [15] and Veatch et al., [45] reported either treatment fidelity and/or social validity data. In terms of

treatment fidelity [42], measured the content of the training workshops, and [21] measured the delivery of the behavioral parent training as intended and parent adherence to training delivered. Both studies received favorable feedback in relation to the parent training provided which demonstrated that the training was standardized, consistent, and effective. Fidelity feedback within the current context could ultimately increase the replication of the training, promote dissemination of the research, and encourage generalization of the training to other settings. Recent research suggests that performance feedback is the most effective way to improve treatment fidelity [8], and that this feedback is crucial to the successful transition of evidence-based interventions into practice. This has implications for the evidence presented in the current review, as only two of the 11 studies reported treatment fidelity data. This has the potential of weakening the identification of the parent training components which may have been either ineffective or effective within behavioral sleep interventions. Future research should attempt to investigate these components and investigate under which conditions parent training obtains optimal outcomes, to consider and understand what motivates parents' behavior and their application of the sleep knowledge obtained from parent training programs.

In contrast, nine of the studies included in the current review received positive social validity feedback; this would suggest that parents found behavioral sleep interventions to be effective, acceptable, supportive, and relevant to their child's sleep problems. Social validity plays a significant role in assisting researchers to generate ongoing evaluation and to implement improvements to the intervention strategies employed. This was a promising outcome; nonetheless, social validity should not be considered as a replacement for the measurement of treatment fidelity.

4.5. Strengths and limitations

It is possible that insufficient inclusion of methodological procedures relating to parent sleep education and behavioral interventions within existing research has contributed to the small number of studies included in the current review. Each study within the current review was examined separately and then compared on common features. Based on the evidence, it is apparent that parent training and professional support have a vital role to play in the success of behavior sleep interventions. As such, future behavioral sleep interventions should consider the additional resources required to provide additional professional support and guidance. Even though the extent of the evidence presented is scant, it nonetheless strengthens the support for the implementation of parent training programs within behavioral sleep interventions. It is also important to clarify that the low level of evidence may be a result of the substantial number of studies excluded based on the exclusion criteria to not include pharmacological interventions rather than the lack of research into the treatment of sleep problems for children with ASD/ID. A further strength of the current review was the inclusion of a range of research designs, diverse sample populations, a variety of parent training programs, and a range of outcome measures to maximize the use of the evidence available. This approach not only enabled an evaluation and comparison of the evidence presented, it also enabled a synthesis of the studies included in the current review. However, this also created a limitation, as the studies included in the review were significantly heterogeneous, and therefore it was not appropriate to conduct a meta-analysis. Depending on the variability within the research, a meta-analysis may be used to combine and to directly compare quantitative results, providing additional information such as the statistical efficacy of parent training programs. As research into parent training and behavioral

sleep interventions for children with ASD/ID advances and more homogeneous studies exist, future research would benefit from the inclusion of a meta-analysis within systematic reviews. Even though the studies in the current review produced results that were too different to combine in a meta-analysis, the formal and rigorous systematic approach enabled a level of comparability of the distinct aspects of information presented within each study and provided an opportunity to assess the consistency of the evidence. It would appear, regardless of the variability within the current studies, that parent training was an effective component of behavioral sleep interventions. The narrative synthesis offered within the current review should be considered with the previously stated limitations and strengths in mind. In addition, the current findings may not generalize to other clinical populations.

5. Conclusion

The current review conveys an emerging evidence-based practice that could contribute to future behavioral sleep research and guide best-practice decisions to support effective parent training to improve sleep outcomes for children with ASD and/or ID. Quality research should be conducted to demonstrate the efficacy and appropriateness of parent training incorporated in behavioral sleep interventions. The evidence presented in the current review may inform and guide future clinical practice in the management of pediatric sleep problems and enable clinicians and families to make informed choices. Future behavioral sleep interventions would benefit from increased availability and accessibility of parent training programs, along with additional professional support to promote meaningful treatment outcomes. It would be beneficial if future studies increased the sample size, as this could have the potential of demonstrating outcomes more clearly with greater statistical power. Future large-scale RCTs could investigate and identify the effective components of parent training programs for children with ASD/ID. It is equally important that future studies incorporate a measurement of treatment fidelity, as this could provide the knowledge required to shed more light on the training content, duration, mode of delivery, level of professional support, and training delivered as intended. Regarding immediate and postintervention effects and maintenance of improved sleep behaviors, researchers should be mindful of the parents' ongoing role of supporting the positive outcomes in the months following treatment. Future research designed to evaluate the immediate and postintervention effects would be of significant value, as analyses of follow-up data enable an understanding of the retention of the outcomes and could be vital to evaluating the overall efficacy of parent training programs. There is a necessity for further evaluation studies, as sleep problems are extremely prevalent and have significant negative consequences for the individual who experience sleep problems and for the families who support them.

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Conflict of interest

The authors have no conflict of interest to declare.

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