Non-pharmacological interventions for non-respiratory sleep disturbance in children with neurodisabilities: a systematic review

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ABBREVIATIONS

CSHQ	Child Sleep Habits Questionnaire
HTA	Health Technology Assessment
NIHR	National Institute for Health Research
RCT	Randomized controlled trial
SOL	Sleep onset latency

AIM To describe existing evidence on non-pharmacological interventions to manage sleep disturbance in children with neurodisabilities.

METHOD We systematically reviewed non-pharmacological interventions aimed at improving non-respiratory sleep disturbance in children with neurodisability. Sixteen databases, grey literature, and reference lists of included papers were searched up to February 2017. Two researchers (B.B., C.M., G.S., A.S., A.P.) undertook screening, data extraction, and quality appraisal.

RESULTS Twenty-five studies were included: 11 randomized controlled trials and 14 before-and-after studies. All studies were at high or unclear risk of bias. Parent-directed interventions were categorized as comprehensive tailored interventions (*n*=9), comprehensive non-tailored interventions (*n*=8), and non-comprehensive interventions (*n*=2). Six 'other' non-pharmacological interventions were included. Seventy-one child and parent sleep-related outcomes were measured across the included studies. We report the two most commonly measured outcomes: the Child Sleep Habits Questionnaire and sleep onset latency. Five studies reported significant improvements on at least one of these outcomes.

INTERPRETATION Various types of non-pharmacological intervention for managing sleep disturbance have been evaluated. Clinical heterogeneity and poor study quality meant we could not draw definitive conclusions on the effectiveness of these interventions. Current clinical guidance recommends parent-directed interventions as the first approach to managing sleep disturbance; prioritizing research in this area is recommended.

Non-respiratory sleep disturbances are more prevalent in children with neurodisabilities than in typically developing children.[1,2] Sleep problems can affect quality of life, school performance, and daytime behaviour.[3,4] Child sleep problems are also associated with poor outcomes for parents and other members of the household.[5]

Current guidance on management of sleep disturbance in children proposes that once clinical or respiratory reasons for sleep disturbance are excluded, interventions that aim to change parents' management of their child's sleep should be the 'first port of call'.[6] This guidance is regarded as applicable to children with neurodisability. Pharmacological interventions (such as melatonin) are recommended where such interventions prove ineffective or alongside parent-directed approaches.[7,8] Other non-pharmacological approaches include chronotherapy, phototherapy, dietary interventions, sensory interventions (e.g. weighted blankets), cranial osteopathy, and environmental changes.

Previous systematic reviews in the field of managing sleep disturbance in children with neurodisabilities have mainly focused on individual diagnoses[9–14] and/or a specific intervention or pharmacological intervention only.[10,13–15] A systematic review was therefore commissioned by the UK National Institute for Health Research (NIHR), Health Technology Assessment (HTA) Programme to collate the existing evidence across multiple interventions and neurodisabilities.

We aimed to assess the effectiveness of non-pharmacological interventions for non-respiratory sleep disturbance in children with neurodisabilities and to identify priorities for future primary research. The review reported here is part of a broader review, which also included pharmacological interventions and will be available as an NIHR HTA

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journal report (https://www.journalslibrary.nihr.ac.uk/programmes/hta/1421202/#/).

METHOD

The review was conducted in accordance with the Centre for Reviews and Dissemination's guidance for undertaking reviews in health care[16] and reported according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines.[17] The review was prospectively registered with PROSPERO (registration number CRD42016034067).[18] As this paper represents a systematic review of published work, ethical approval was not required.

Eligibility criteria

Studies were assessed against the eligibility criteria described in the following sections.

Population

Children and young people (0–18y) with neurodisability and experiencing non-respiratory sleep disturbance were included. Neurodisability was defined in accordance with the consensus definition of Morris et al.[19] Non-respiratory sleep disturbances of any duration relating to initiation, maintenance, or scheduling of sleep, diagnosed by a health care professional on the basis of parental/carer or child report or sleep observation, were included. Central disorders of hypersomnolence and sleep-related movement disorders were excluded.

Intervention

Non-pharmacological interventions aimed at improving sleep initiation, maintenance, scheduling, or quality in any setting, which are relevant to the care provided by statutory health care services across the UK, were included. Interventions had to meet current practice standards[20] on the basis of guidance from clinical members of the team (e.g. interventions that involved punishment were not eligible).

Comparator(s)

Studies using no comparator, wait list control, placebo, or other active intervention were eligible.

Outcomes

The primary outcomes of interest were child- and parent-related sleep. These included actigraphy-based and parent/carer- or child-reported measures (e.g. sleep diaries, or standardized scales relating to initiation, maintenance, scheduling, or quality of sleep).

Secondary outcomes included child-related quality of life; daytime behaviour and cognition; parent/carer quality of life and well-being including global quality of life, physical well-being, mental well-being, mental health (e.g. stress) and family functioning; and adverse events.

Study design

Randomized controlled trials (RCTs), non-randomized controlled studies, and before-and-after studies were eligible. Case studies were excluded.

What this paper adds

- Existing evidence on non-pharmacological interventions to manage sleep disturbance in children with neurodisabilities is predominately of poor quality.
- Most included studies evaluated parent-directed interventions of varying content and intensity.
- There was very little consistency between studies in the outcome measures used.
- There is some evidence that parent-directed interventions may improve child outcomes.

Search strategy

An information specialist searched the following electronic databases in February and March 2016 and updated the search in February 2017: Applied Social Sciences Index of Abstracts (ASSIA); The Cochrane Central Register of Controlled Trials (CENTRAL); Cochrane Database of Systematic Reviews; Conference Proceedings Citation Index; Cumulative Index of Nursing and Allied Health Literature (CINAHL); Database of Abstracts of Reviews of Effects; Embase; Health Management Information Consortium; MEDLINE; MEDLINE In-Process; PsycINFO; Science Citation Index; Social Care Online; and Social Policy & Practice. ClinicalTrials.gov; World Health Organization International Clinical Trials Registry Platform; and the UK Clinical Trials Gateway were also searched for ongoing and completed trials. An example search strategy (for ASSIA) is provided in Appendix S1 (online supporting information). There were no restrictions on date, language, or study design.

Study selection and data extraction

The search results were downloaded into Endnote bibliographic software (Clarivate Analytics, Philadelphia, PA, USA) and deduplicated. The first 10% of titles were screened independently by two researchers (B.B., C.M., G.S., A.S., A.P.). Once agreement had been reached, a single researcher (A.S., A.P.) screened the remainder. Two researchers (B.B., C.M., G.S., A.S., A.P.) independently screened the abstracts of the records identified as potentially relevant on the basis of their title. Full papers were independently screened by two researchers (B.B., C.M., G.S., A.S., A.P.). Discrepancies were resolved through discussion and consensus with a third researcher (C.M.) if necessary. Data extraction forms were developed and piloted in Microsoft Word 2010 and Excel 2010. Data extracted included details of study design, descriptions of the intervention and comparator, outcome measures, and methods of assessment. Outcome data were extracted to allow calculation of the mean difference and 95% confidence interval (CI) between groups to assist comparison between studies. Data extraction was undertaken by one researcher and checked by a second (A.S., A.P.).

Quality assessment

Risk of bias was assessed using the Cochrane Risk Of Bias Tool for RCTs,[21] A Cochrane Risk Of Bias Assessment Tool for Non-Randomized Studies of Interventions for other studies with a control group,[22,23] or an adapted

checklist for before-and-after studies.[24] For crossover trials, we also assessed whether an appropriate analysis using paired data was conducted and whether there was a treatment-by-period interaction.[25] Assessment of risk of bias was undertaken independently by two researchers (B.B., C.M., G.S., A.S., A.P.), with discrepancies resolved through consensus, or discussion with a third researcher (C.M.).

Strategy for data synthesis

The substantial heterogeneity of interventions, study design, and outcome measures across studies meant meta-analysis was not appropriate. Therefore, narrative summaries are used to describe the available evidence. Interventions were assigned to the following categories: parent-directed and 'other' non-pharmacological interventions (Appendix S2, online supporting information).

Parent-directed interventions were defined as psychoeducational interventions aiming to teach parents knowledge and skills to manage their child's sleep disturbance and possibly to provide support to parents as they implement new knowledge and skills. Modes of delivering such interventions include one-to-one sessions, group work, one-off workshops, and provision of written material. Given the variety within this category of intervention, these were classified in terms of their content (comprehensive vs non-comprehensive) and the degree to which they were personalized to the individual child (tailored vs non-tailored). The following intervention typology was created:

(1) Comprehensive tailored: a detailed assessment guides the decision-making regarding the management of a specific child's sleep disturbance. A sleep management plan specific to the child/family is developed, and training in implementing that plan is delivered. There is ongoing support and advice as parents implement changes to sleep management strategies and practices ('implementation support'). A comprehensive approach is used involving training across sleep and sleep processes, sleep hygiene, and the management of specific problem behaviours (e.g. night wakings).
(2) Comprehensive non-tailored: a standard 'training curriculum' is used which is comprehensive in content and may include opportunities for a parent to be supported to operationalize the material learnt to their child's sleep disturbance. Implementation support may also be included.
(3) Non-comprehensive: intervention focuses on a single topic area related to managing sleep disturbance (e.g. sleep hygiene, behavioural strategies); these may be tailored or non-tailored.

Other types of non-pharmacological intervention included interventions such as complementary therapies and weighted blankets.

Studies were grouped by intervention type, study objective (evaluations of intervention effectiveness, evaluations of different modes of delivering an intervention or intensity of support), and then by study design (RCT and non-randomized study designs) for the synthesis.

RESULTS

Overview of the evidence

After deduplication, 15 745 titles were screened and 25 studies investigating non-pharmacological interventions were included (Fig. 1). A list of excluded studies is available from the authors.

Table I summarizes key study characteristics, grouped by the type of intervention evaluated. Eleven RCTs, one controlled before-and-after study, and 14 uncontrolled before-and-after studies were included. Studies were conducted in the UK (n=9), USA (n=7), Australia (n=5), and one each in Canada, Hong Kong, Israel, and China. Sample sizes ranged from 5 to 244 participants.

The mean age of children ranged from 2 years 8 months to 12 years 1 month. Thirteen studies included children with two or more neurodisabilities. In nine studies, participants were described as having a single neurodisability diagnosis: autism spectrum disorder (n=6) or attention-deficit–hyperactivity disorder (ADHD) (n=3). The remaining three studies offered no detail on the types of neurodisability represented; generic terms such as 'mental retardation' were used. Most studies included children with a mix of sleep disturbances, with the most commonly reported being sleep initiation and maintenance (n=14 studies). The first timepoint at which outcomes were measured once the intervention was completed ranged from immediately after intervention to 2 months after intervention. Five trials collected outcome data at additional follow-up time points; however, to minimize heterogeneity in results we only report outcomes measured closest to the end of the intervention.

Risk of bias

Poor reporting of study methods and results was found across all study designs. All RCTs were assessed as having high risk of bias for most items on the Cochrane Risk of Bias tool because of issues with randomization and incomplete outcome data. We were unable to find a registered protocol for 10 RCTs,[5,26–34] and in all RCTs blinded outcome assessment was either not undertaken or it was unclear whether blinding had occurred.[5,26–32,34–36] However, we do note that the type of interventions and outcomes under investigation make robust, blinded outcome assessment challenging. Although the use of actigraphy data may be considered more objective than parent-reported data in terms of the measurement of some sleep outcomes, we did not consider these to be true objective outcomes with non-blinding likely to introduce bias.

Non-randomized studies were at high (n=12) or unclear (n=2) risk of bias. This was mainly because of how studies selected participants (e.g. not reporting eligibility criteria)[37–50] and likely or unclear bias in measurement of intervention outcomes.[39,40,43–46,51]

Outcomes

Seventy-one sleep-related outcomes were reported across the included studies. Given the number of outcomes

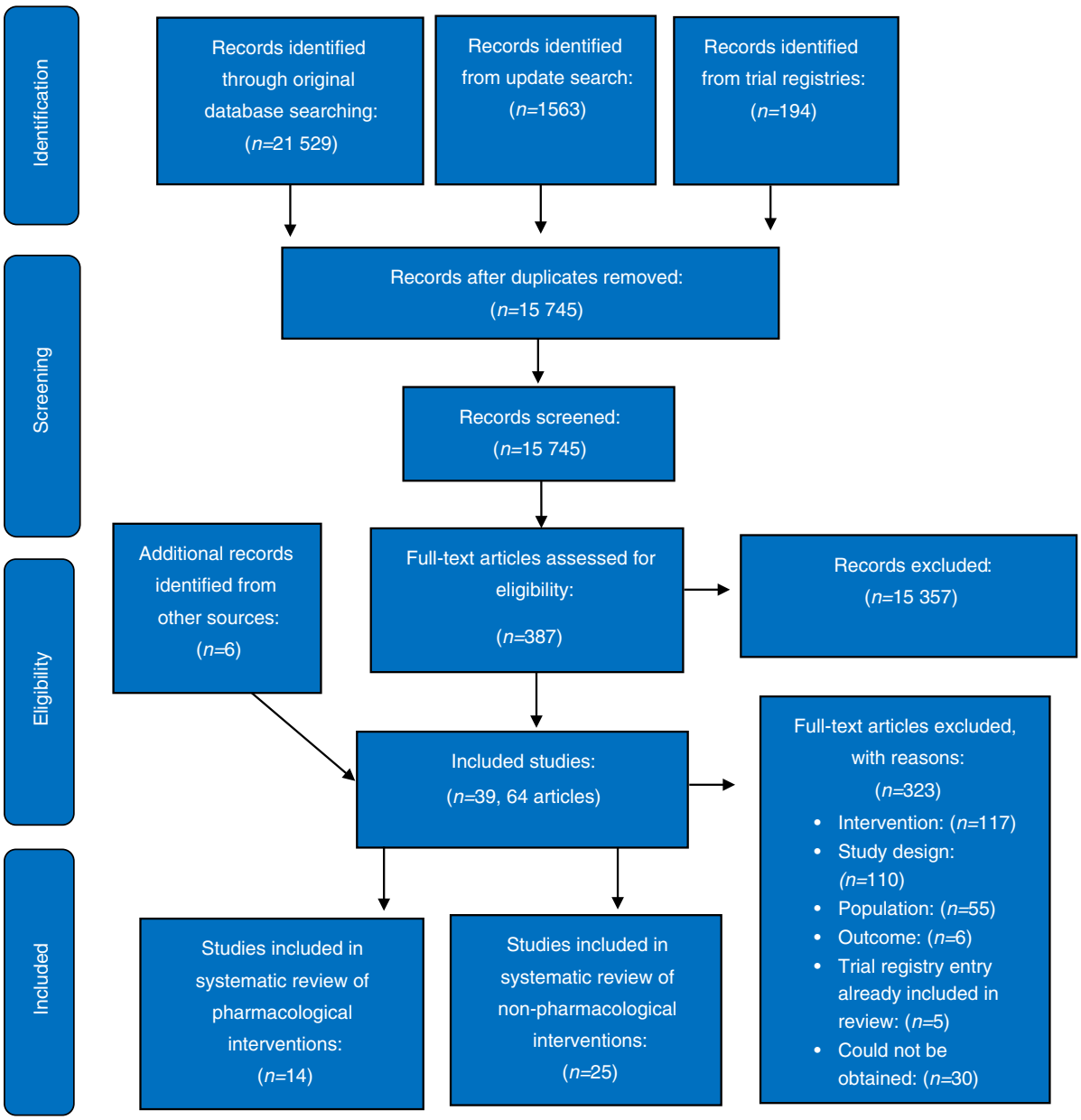

Figure 1: Flow chart of the study selection process. [Colour figure can be viewed at wileyonlinelibrary.com].

assessed, in this paper we only report the two most commonly measured outcomes: Child Sleep Habits Questionnaire (CSHQ),[52] and sleep onset latency (SOL). The CSHQ is a parent-report questionnaire which is widely used to measure sleep disturbance. The questionnaire has 33 items, rated on a 3-point Likert scale. Items are grouped into the following subscales: bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night wakings, parasomnias, sleep disordered breathing, and daytime sleepiness. A total score offers an overall measure of sleep disturbance, with higher scores indicating a greater severity of sleep disturbance, owing to either the frequency (i.e. regularity) or number of different behaviours presenting. However, caution is needed when using the scale as the sole method of assessing a child's sleep problems as a number of subscales showed low construct validity and diagnostic validity.[53] No clinically important difference has been established for either the CSHQ or SOL. At least one of these outcomes was reported by most included studies. Six studies did not report either of these outcomes.[26,34,36,37,43,46] Full data on all outcomes are provided in the HTA report (https://www.journalslibrary.nihr.ac.uk/programmes/hta/1421202/#/).

Results from studies

Parent-directed: comprehensive tailored interventions

Five RCTs[5,27–29,35] and four before-and-after studies[5,43,50,51] evaluated comprehensive tailored interventions,

Table I: Study characteristics

Main publication (associated papers) Country	Study design Participants randomized (total *n* and by group) Intervention (I) Comparator (C)	Intervention (I) Comparator (C)	Mean age (SD), y:mo Neurodisability disorder	Sleep disturbance	Risk of bias
Comprehensive tailored interventions					
Austin et al.[50] Australia	Before-and-after study *n*=8	I: Two training workshops, home visit for assessment, development of sleep management strategy, then third workshop. Implementation support by weekly telephone call	4:0 (1:11) Mixed	Sleep initiation and maintenance	High
Beresford et al.[5] Associated papers[64–66] UK	Parallel group RCT *n*=13 I: *n*=7 C: *n*=6	I: Two face-to-face sessions for assessment, development of sleep management and parent training strategies. Telephone implementation support C: Usual approach to providing SMI: as above, but implementation support by home visit	I: 2:10 (0:10) C: 2:8 (1:00) Mixed	Sleep initiation and maintenance	High
Beresford et al.[5] intervention 2 Associated papers[64–66] UK	Before-and-after study *n*=12	I: Two assessment sessions, development of sleep management strategy, and training parent in strategy. Fortnightly face-to-face implementation support	2:11 (1:35) Mixed	Sleep initiation and maintenance	High
Hiscock et al.[35] Associated paper[67] Australia	Parallel group RCT *n*=244 I: *n*=122 C: *n*=122	I: One assessment session, development of sleep management and parent training strategies. Implementation support by one face-to-face session and one telephone call C: Usual care: routine access to health care from paediatrician, where required. Sleep issues not routinely addressed	I: 10:4 (1:10) C: 9:11 (2:1) ADHD+learning disability or ASD/Asperger syndrome	Sleep initiation	High
Johnson et al.[27] Associated paper[68] USA	Parallel group RCT *n*=40 I: *n*=20 C: *n*=20	I: One assessment session, development of sleep management strategy, five sessions training parent in strategy. Face-to-face implementation support C: Non-sleep-related parent education delivered in identical manner to intervention group	I: 3:6 (1:00) C: 3:7 (1:1) Autism and ASD	Sleep initiation and maintenance	High
Moss et al.[28] Associated papers[68,69] Australia	Parallel group RCT *n*=26 I: *n*=13 C: *n*=13	I: Two training workshops, home visit for assessment, and development of sleep management strategy. Implementation support by home visit and telephone calls as required. C: Waiting list control	11:9 (2:6) (not reported separately) Mixed	Sleep initiation, maintenance, and scheduling, and snoring	High
Quine and Wade[43] Associated paper[70] UK	Before-and-after study *n*=25	I: Two assessment sessions, development of sleep management strategy, and training parent in strategy. Face-to-face implementation support	Mean (SD) not reported (range 3–21y) Learning disability	Sleep initiation and maintenance	High
Sciberras et al.[29] Associated papers[70–72] Australia	Parallel group RCT *n*=27 I: *n*=14 C: *n*=13	I: Two assessment sessions, development of sleep management strategy, training parent in strategy. Implementation support by telephone call and face-to-face visit if needed C: Single assessment session, development of sleep management strategy, and training parent in strategy. No implementation support	I: 12:1 (2:2) C: 10:11 (2:6) ADHD	Sleep initiation	High

Table I: Continued

Main publication (associated papers) Country	Study design Participants randomized (total *n* and by group) Intervention (I) Comparator (C)	Intervention (I) Comparator (C)	Mean age (SD), y:mo Neurodisability disorder	Sleep disturbance	Risk of bias
Weiskop et al.[51] Australia	Before-and-after study *n*=13	I: Four assessment sessions, development of sleep management strategy, and training parent in strategy. Telephone implementation support delivered from start of intervention and continued until after training sessions finished with a face-to-face session and further telephone calls	5:1 (2:0) Mixed	Sleep initiation and maintenance	High
Comprehensive non-tailored interventions					
Adkins et al.[30] Associated paper[70] USA	Parallel group RCT *n*=36 I: *n*=18 C: *n*=18	I: Training curriculum in a booklet given to parent C: No booklet provided	6:5 (2:7) (not reported separately) Mixed	Sleep initiation	High
Beresford et al.[5] intervention 3 Associated papers[64–66] UK	Before-and-after study *n*=22	I: Four sessions, group delivery of training curriculum	8:11 (3:3) Mixed	Sleep initiation	High
Beresford et al.[5] intervention 4 Associated papers[64–66] UK	Before-and-after study *n*=25	I: Training curriculum delivered by single half-day workshop	7:0 (3:4) Mixed	Sleep initiation and maintenance	High
Bramble[44] Associated paper[73] UK	Before-and-after study *n*=15	I: Training curriculum by single session. Implementation support by telephone calls	7:2 (2:7) Mixed	Sleep initiation and maintenance	High
Malow et al.[31] USA	Parallel group RCT *n*=80 I: *n*=39 C: *n*=41	I: Training curriculum delivered by two group sessions. Implementation support by telephone calls C: Training curriculum delivered by single face-to-face session. Implementation support delivered by telephone calls	I: 5:11 (2.8) C: 5:7 (2.6) Mixed	Sleep initiation	High
Montgomery et al.[26] Associated paper[26] UK	Parallel group RCT *n*=82 Ia: *n*=22 Ib: *n*=34 C: *n*=26	Ia: Training curriculum contained in a booklet given to parent Ib: Training curriculum identical to that in booklet delivered face-to-face C: Waiting list	Mean (SD) not reported (range 27–101mo) (not reported separately) Mixed	Sleep initiation and maintenance	High
Reed et al.[48] Associated paper[74] Canada	Before-and-after study *n*=22	I: Group delivery of training curriculum over three sessions	5:10 (2:8) ASD	Sleep initiation and maintenance	Unclear
Yu et al.[38] Hong Kong	Before-and-after study *n*=54	I: Group delivery of training curriculum over three sessions, supported by weekly telephone calls. Implementation support by telephone	4.78y (0.85) ASD and Asperger syndrome	Sleep initiation and maintenance	High
Non-comprehensive interventions					
Peppers et al.[47] USA	Before-and-after study *n*=23	I: Prescriptive sleep hygiene intervention. One session via practitioner	Mean (SD) not reported (range 5–11y) Neurodisability not reported	Global measures of sleep disturbance (Child Sleep Habits Questionnaire) used to define eligibility to receive intervention	High

Table I: Continued

Main publication (associated papers) Country	Study design Participants randomized (total *n* and by group) Intervention (I) Comparator (C)	Intervention (I) Comparator (C)	Mean age (SD), y:mo Neurodisability disorder	Sleep disturbance	Risk of bias
Wiggs and Stores[34] Associated papers[73,75] UK	Cluster RCT *n*=30 I: *n*=15 C: *n*=15	I: Tailored intervention, single session for assessment, development of sleep management strategy, training parent in strategy. Implementation support by telephone	I: 8:2 (2:8) C: 10:9 (3:10) Mixed	Sleep initiation and maintenance	High
Other non-pharmacological interventions					
Gringras et al.[32] UK	Crossover RCT *n*=73	I: weighted blanket 2.25kg (small) 4.5kg (large), 12–16d, given by researchers at home/clinic visits C: placebo blanket	Weighted blanket first: 8:8 (3:4) Control blanket first: 9:11 (2:10) Mixed	Sleep initiation and maintenance	High
Guilleminault et al.[37] USA	Before-and-after study *n*=14	I: Light therapy and behavioural programme. Daily light exposure at 07:00 and 12:00	2:11; range 9mo to 4y Moderate to severe intellectual disability	Sleep maintenance and lack of sleep consolidation	High
Oriel et al.[40] USA	A–B–A withdrawal design *n*=8	I: Aquatic exercise programme: 60min of aquatic exercise two times per week	8:11 (SD not reported range 6–11y) ASD	Parent/guardian report of sleep dysfunction	High
Piazza et al.[36] USA	Parallel group RCT *n*=14 I: *n*=7 C: *n*=7	I: Faded bedtime with response costs, 10d. Study author delivered face-to-face home visits and booklet intervention C: Bedtime scheduling, consistent sleep and wake time, and prevention of daytime sleep	I: 6:8 (2:7) C: 8:4 (3:0) Mixed	Sleep initiation and maintenance	High
Yehuda et al.[46] Israel	Controlled before-and-after study *n*=78 I: *n*=40 C: *n*=38 (Healthy control *n*=22 not included)	I: Essential fatty acid supplement, 90g α-linolenic and 160g of linoleic acid in mineral oil. Two capsules per day for 10wks C: Placebo	Mean (SD) not reported (range 9–12y) ADHD	Sleep deprived	Unclear
Yu and Hong[39] China	Before-and-after study *n*=30	I: Acupuncture and ear point taping. Two courses of acupuncture treatment, once every other day, three times a week with 36 sessions constituting one course. Ear point taping three times per week, 36 sessions constituted one course. Two courses required	6:11 (3:1) 'Mental retardation'	Sleep initiation, maintenance, and abnormal sleep (including apnoea)	High

ADHD, attention-deficit–hyperactivity disorder; ASD, autism spectrum disorder; C, comparator; I, intervention; RCT, randomized controlled trial; SMI, sleep management intervention.

delivered face-to-face (at home and/or in clinic) (Table II). The duration of the intervention, the number of sessions delivered, and the extent of implementation support varied across studies.

Of the five RCTs, three used a no-intervention comparator,[27,28,35] and two evaluated alternative ways of delivering an intervention: one compared the mode of implementation support (home visit vs telephone call);[5] and the other compared the intensity of practitioner involvement when delivering the intervention (brief vs extended).[29]

CSHQ. Four RCTs (n=310)[5,28,29,35] and two before-and-after studies (n=20)[5,50] reported the CSHQ total score, a validated parent-reported global assessment of child sleep (Table III). One RCT, which was classified as having low risk of bias on all domains except for performance bias (n=244), reported a statistically significant reduction (i.e. improvement) in total CSHQ score after intervention for the ADHD-specific intervention compared with usual care (adjusted mean difference −6.6, 95% CI: −8.5 to −4.6).[35] Another smaller RCT (n=26) reported a similar magnitude of effect but was not statistically significant (mean difference −4.62, 95% CI: −10.83 to 1.59).[28] In one before-and-after study there was an improvement in total CSHQ score after intervention compared with preintervention (mean difference −7.9, 95% CI: −14.4 to −1.3).[50]

For the two trials investigating alternative approaches to delivering the intervention, no statistically significant difference in CSHQ score was observed.[5,29]

SOL. One RCT (n=40)[27] and two before-and-after studies (n=21)[50,51] measured SOL, the time from bedtime to sleep onset. There was no statistically significant difference in actigraphy-measured SOL (verified using sleep diaries)

Table II: Details of comprehensive tailored interventions (active arms only)

Study	Total duration of intervention (including implementation support)	Mode of delivery of assessment and parent training (excluding implementation support) Number of sessions (location)	Mode of delivering implementation support, and intensity, once regular sessions with practitioner completed	Intervention developed for specific neurodisability?	Manual?	Length of follow-up
Randomized controlled trials						
Beresford et al.[5]	10wks	Face-to-face. One (home)	Home visit: approximately weekly for 6–8wks. Versus telephone call: approximately weekly for 6–8wks	No	No	10wks, 22wks
Hiscock et al.[35]	4wks	Face-to-face. One (home or clinic)	Face-to-face (n=1), later followed by telephone call (n=1)	Yes, attention-deficit–hyperactivity disorder	No	3mo, 6mo
Johnson et al.[27]	Not reported	Face-to-face. Five (home and clinic)	Face-to-face (n=1)	Yes, autism spectrum disorder	Yes	1mo, 2mo
Moss et al.[28]	15wks	Teaching workshops and face-to-face. Two workshops and one face-to-face (home)	Home visit (n=1), followed by telephone calls, 'on a needs basis for approximately 2mo'	No	Yes	15wks, 23wks
Sciberras et al.[29]	One session vs 4wks	Face-to-face. One (clinic) vs two (clinic)	None Versus telephone call (n=1) followed by face-to-face session (clinic) if needed	Yes, attention-deficit–hyperactivity disorder	No	2mo
Before-and-after studies						
Austin et al.[50]	15wks	Teaching workshops and face-to-face. Two workshops and one home visit and one workshop	Approximately weekly telephone call for 6wk period	No	Yes	19wks
Beresford et al.[5]	12–16wks	Face-to-face. Two (clinic, home)	Fortnightly sessions at clinic	No	No	12wks, 24wks
Quine and Wade[43]	6–28wks	Face-to-face. Two (home)	Described as 'weekly' home visits, although study authors also report frequency decided between practitioner and parent and diminishing in intensity	No	Yes	3mo
Weiskop et al.[51]	Minimum 7wks	Face-to-face. Four (mix of home and clinic), plus at least weekly telephone contact between sessions	'Review session' 5wks after session 4; telephone calls 'gradually reduced' after session 5	No	Yes	3mo, 12mo

Table III: Outcome results for Child Sleep Habits Questionnaire for comprehensive tailored interventions

Study	Baseline, mean (SD)	Follow-up, mean (SD)	Mean difference[a] (95% CI)
Randomized controlled trials			
Beresford et al.[5]	I: 59.50 (11.82)	I: 52.17 (11.44)	−1.16 (−14.27 to 1.95)[a]
	C: 53.33 (4.27)	C: 53.33 (8.76)	
Hiscock et al.[35]	I: 57.8 (8.8)	I: 50.1 (8.3)	Adjusted: −6.6 (−8.5 to −4.6)[b]
	C: 59.0 (7.8)	C: 55.1 (8.6)	−5.0 (−7.6 to −2.4)[a]
Moss et al.[28]	I: 56.20 (9.38)	I: 46.50 (7.29)	−4.62 (−10.83 to 1.59)[a]
	C: 51.38 (7.54)	C: 51.12 (6.51)	
Sciberras et al.[29]	NR	I: (change score) 5.09 (5.12)	−1.73 (−7.11 to 3.65)[c]
		C: (change score) 6.82 (8.02)	
Before-and-after studies			
Austin et al.[50]	55.43 (7.68)	47.57 (9.14)	−7.86 (−14.39 to −1.33)[a]
Beresford et al.[5]	59.55 (7.59)	56.57 (10.77)	Cannot be estimated.[d] Effect size given as 0.42.

[a]Unadjusted mean difference unless otherwise stated. [b]Reported in paper. [c]Difference in change scores from baseline to 2mo between intervention and control groups. [d]As not a matched sample (n=11 preintervention and n=7 postintervention). I, intervention; C, comparator; NR, Not reported

in the RCT of a comprehensive tailored intervention compared with an attention placebo control (non-sleep-related parent education) (mean difference 4min, 95% CI: −15.0 to 23.0).[27] One before and after the study also reported no statistically significant difference before and after the intervention in sleep-diary-measured SOL (mean difference 43min, 95% CI: −30 to 116);[50] the second presented the results as graphs with no numerical data available.[51]

Parent-directed: comprehensive non-tailored interventions

Three RCTs and five before-and-after studies evaluated comprehensive non-tailored interventions.[26,30,31,38,44,48,54] Various modes of delivery were used across the studies (Table IV). They also varied in the extent to which they accommodated the specific information and training needs parents might have had for their child's condition and/or sleep problem. Some included telephone implementation support, whereas others did not.

One RCT compared a sleep training curriculum delivered by a booklet with no intervention;[30] one compared two modes of delivering the same curriculum group versus individual face-to-face sessions supplemented by weekly telephone calls;[31] and one compared group with individual delivery of a training curriculum.[26] The before-and-after studies evaluated a group-delivered intervention;[38,42,48] a single session workshop;[41] and an individually delivered intervention.[44]

CSHQ. One RCT (n=80)[31] and four before-and-after studies (n=126)[38,41,42,48] reported CSHQ total score. The RCT reported no statistically significant difference for this outcome between delivery of the training curriculum via a group or a single face-to-face session (not possible to calculate effect estimate and 95% CI).[31] Two before-and-after studies, one evaluating a three-session group-delivered intervention[48] and the other a four-session group-delivered intervention plus implementation support,[38] reported statistically significant improvements (i.e. a decrease) in CSHQ total score after intervention compared with preintervention (mean difference −6.9, 95% CI: −2.6 to −11.2[48] and mean difference −3.3, 95% CI: −1.4 to −5.3[38] respectively). For the two other before-and-after studies, the mean difference in total CSHQ score could not be calculated before and after the intervention as the samples were not matched. However, the studies reported small or very small effect sizes of 0.20 and 0.02.[41]

SOL. Two RCTs (n=116)[30,31] and two before-and-after studies (n=40)[44,48] reported SOL. No statistically significant difference in SOL was observed in the RCT comparing a non-tailored intervention with no intervention (mean difference −11.8, 95% CI: −37.3 to 13.7),[30] the RCT comparing individual versus group delivery of the same training curriculum (mean difference −0.2, 95% CI: −9.9 to 9.5),[31] or in the before-and-after study of a group-delivered intervention (data not presented, narrative report provided only).[48] The second before-and-after study reported a statistically significant reduction in SOL after receipt of a non-tailored comprehensive intervention delivered by a single face-to-face session (mean difference −42.8, 95% CI: −6.01 to −24.6).[44]

Parent-directed: non-comprehensive interventions

One RCT and one before-and-after study[34,47] evaluated non-comprehensive interventions (Table V).

The RCT (n=30) evaluated an intervention that focused specifically on behavioural principles of managing problem sleep.[34] The comparator was an attention control. Neither CSHQ nor SOL were reported in this study. The before-and-after study (n=23) evaluated an intervention[47] that trained parents of children with ADHD on the principles of sleep hygiene only. This study reported a statistically significant improvement in CSHQ total score at 6 weeks after intervention (mean difference 6.4, 95% CI: 4.3–8.5).

Other non-pharmacological interventions

Two RCTs and four before-and-after studies evaluated other types of non-pharmacological intervention (Tables I and VI).[32,36,37,39,40,46]

CSHQ. One study reported the CSHQ; there was a statistically significant reduction in total CSHQ score in the before-and-after study after acupuncture and ear point taping (mean difference −11.5, 95% CI: −13.3 to −9.7).[39]

Table IV: Details of comprehensive non-tailored interventions (active arms only)

Study	Total duration of intervention (including period of implementation support)	Mode of delivery	Number of sessions over which curriculum delivered	Opportunity to operationalize curriculum content to child's sleep problem	Mode of delivering implementation support, and intensity, once curriculum delivered: mode and intensity	Intervention developed for specific neurodisability?	Manual	Length of follow-up
Randomized controlled trials								
Adkins et al.[30]	N/A	Booklet	N/A	No	None.	Yes, autism spectrum disorder	N/A	2wks
Malow et al.[31]	2wks	Face-to-face vs Group	One vs two	Yes	Weekly telephone call (*n*=2) after sessions completed	Yes, autism spectrum disorder	Yes	1mo
Montgomery et al.[26]	N/A vs one session	Booklet vs Face-to-face	N/A vs one	No vs no	None vs none	No	Yes	6wks
Before-and-after studies								
Beresford et al.[42] intervention 3	5wks	Group	Four	Yes	None (but included within curriculum for group session)	No	Yes	5wks, 17wks, 29wks
Beresford et al.[41] intervention 4	One session	Teaching workshop	One	No	None	No	Yes	12wks, 24wks
Bramble[44]	One session	Face-to-face (clinic)	One	Minimal ('only minor individual tailoring')	Telephone calls on three consecutive days after session. Additional calls arranged if necessary	No	Yes	2wks, 4mo, 18mo
Reed et al.[48]	3wks	Group	Three	Yes	None (but included within curriculum for group sessions)	Yes, autism spectrum disorder	Yes	7wks
Yu et al.[38]	7wks	Group, plus weekly telephone calls	Three	Yes	Weekly for 4wks	Yes, autism spectrum disorder	Yes	3wks, 7wks, 11wks

Table V: Details of non-comprehensive interventions (active arms only)

Study design	Intervention content	Total duration of intervention	Mode of delivery	Number of sessions and location	Mode of delivering implementation support, and intensity, once regular sessions with practitioner completed		Intervention described as developed for specific neurodisability?	Manual	Length of follow-up
Wiggs and Stores[34] Randomized controlled trial	Behavioural principles of managing problem sleep behaviour	Unclear	Face-to-face (home)	One (home)	Weekly phone calls. Continued for at least a month, total duration unclear		No	Yes	Postintervention at 'visit 4' approximately 1mo after randomization and 'visit 6' approximately 3mo after randomization
Study design	Intervention content	Total duration of intervention	Mode of delivery	No sessions over which curriculum delivered	Opportunity to operationalize curriculum content to child's sleep problem?	Mode of delivering implementation support, and intensity, once curriculum delivered: mode and intensity	Intervention described as developed for specific neurodisability?	Manual?	Length of follow-up
Peppers et al.[47] Before-and-after study	Principles of sleep hygiene for children with attention-deficit–hyperactivity disorder	One session	Face-to-face (clinic)	One (clinic)	Yes	None	Yes, attention-deficit–hyperactivity disorder	Unclear	6wks

Table VI: Details of other non-pharmacological interventions (active arms only)

Study	Type of intervention	Details of intervention	Total duration of intervention. Mode of delivery and location	Intervention described as developed for specific neuro-disability?	Length of follow-up
Randomized controlled trials					
Gringras et al.[32]	Weighted blanket	During a home or clinic visit, children were given a weighted blanket at baseline and used for 12–16d. Blanket weighed 2.25kg (small) 4.5kg (large). Additional clinic/home visit	Participants used the blanket for 12–16d Blanket received at home or clinic	No	4wks
Piazza et al.[36]	Faded bedtime and response costs	Faded bedtime with response cost involved establishing a bed time where it was likely the child would fall asleep within 15min. Response cost involved keeping the child awake for 1h if they did not fall asleep within 15min of bedtime	'Average treatment length 8wks'. Face-to-face (hospital)	No	'After 10d of an average 8wks treatment'
Before-and-after studies					
Guilleminault et al.[37]	Light therapy and behavioural programme	Light therapy and behavioural programme. Children were exposed to bright light (sunlight or artificial). The behavioural programme involved scheduled parent child interaction; scheduled naps for younger children; avoidance of naps for older children; scheduled lunch; scheduled sleep time	Unclear	No	6mo
Oriel et al.[40]	Aquatic exercise programme	Aquatic exercise programme. During all three phases of the study, the researchers made telephone calls to parents/guardians questioning them about their child's previous night of sleep (twice a week). The programme consisted of warm-up exercises; upper and lower extremity circuits; cardiovascular exercises; a game, which included red light–green light, keep away, or sharks and minnows; free swim in which the participants were given the opportunity to play with toys; and cool-down. Participants were continuously encouraged to remain active throughout the entire session	60min of aquatic exercise two times a week Unclear	No	4mo, 8mo, and 12mo from start of intervention
Yehuda et al.[46]	Dietary intervention	Essential fatty acid supplement, which comprised 90g α-linolenic acid and 360g of linoleic acid in mineral oil	Two capsules for 10wks Unclear	No	10wks

Table VI: Continued

Study	Type of intervention	Details of intervention	Total duration of intervention. Mode of delivery and location	Intervention described as developed for specific neuro-disability?	Length of follow-up
Yu and Hong[39]	Alternative therapy	Acupuncture and ear point taping (see full paper for technical details of acupuncture and ear point taping)	Two courses of acupuncture treatment were given once every other day, three times a week, with 36 sessions constituting one course Ear point taping was given three times a week, with 36 sessions constituting one course. Two courses were required Unclear	No	'After treatment'

SOL. One RCT (n=73)[32] and one before-and-after study (n=8)[40] measured SOL. There was no statistically significant difference for this outcome in the RCT comparing weighted blankets with placebo blankets[32] (actigraphy-measured SOL: mean difference 2.1, 95% CI: −5.5 to 9.7; parent reported: mean difference −1.6, 95% CI: −6.7 to 3.5). There was also no statistically significant difference in parent-reported SOL in a before-and-after study evaluating an aquatic exercise intervention (mean difference 19.11, 95% CI: −40.95 to 6.57).[40]

DISCUSSION

Principal findings

This systematic review has identified a lack of high-quality evidence assessing the effectiveness of non-pharmacological interventions to manage sleep disturbance in children with neurodisabilities.

Three-quarters of the studies evaluated parent-directed interventions. We found no replication of studies or more than one study evaluating the same intervention. This lack of evidence is noteworthy given that parent-directed interventions are recommended as the 'first port of call' for clinicians seeking to manage sleep disturbance in children with neurodisabilities. Less than half the evidence came from RCTs, and all of these had substantial or unclear risk of bias; therefore their findings need to be treated with some caution.

Several of the parent-directed interventions showed evidence of benefit. One before-and-after study reported a significant reduction in SOL after a comprehensive non-tailored intervention delivered by a single face-to-face session.[44] In relation to total CSHQ score, one RCT of a comprehensive tailored intervention developed specifically for children with ADHD, and rated as being at low risk of bias for all domains except blinding,[35] reported a statistically significant improvement on this outcome measure. Additionally, two before-and-after studies evaluating comprehensive non-tailored interventions delivered via groups,[38,48] and one before-and-after study of a non-comprehensive intervention (ADHD-specific) also showed statistically significant improvements in total CSHQ score.[47] As far as we are aware, a minimum clinically important difference for the CSHQ has not yet been established; therefore the clinical significance of these findings is unclear. We would note that where RCTs did not show evidence of statistically significant benefit for outcomes, this too needs to be interpreted cautiously as 'no evidence of effect' does not mean there is 'evidence of no effect'.[54]

For the RCTs that were comparing parent-directed interventions in terms of mode of delivery or intensity of support, the evaluation question is different and is concerned with comparing the effectiveness of alternative (in the case of this review, more and less resource intensive) ways of providing an intervention.[5,27,29,31] In three of these trials,[5,29,31] no significant differences in outcomes, as assessed by CHSQ scores and/or SOL, were reported. The remaining trial[27] did not use these outcome measures. Again, we reiterate the

caution with which these findings should be treated given the reported issues with study quality, the lack of any replication, and the absence of a rubric by which the clinical significance of observed effects can be judged.

Comparison with other research

Our results support the findings of Brown et al.,[55] who, in 2013, reviewed evidence on non-pharmacological interventions to manage sleep disturbance in children with neurodevelopmental disorders, or cognitive and/or visual impairment. Their preliminary scoping searches suggested that most studies, particularly those with a more robust design, had not yet reported and so it was considered too early to attempt a systematic review of RCTs. Instead, the authors conducted a critical review and concluded that there is little conclusive evidence on non-pharmacological interventions in this population. In contrast, a recent systematic review of parent-directed sleep management interventions for non-disabled children aged 5 years and under concluded that there was 'moderate support' for these interventions. The authors recommended parent-directed interventions to be implemented without hesitation for typically developing young children.[56]

Existing evidence on non-pharmacological interventions for sleep disturbance in children with neurodisabilities provides little clarity as to the effectiveness of these interventions. Given the poor quality and inconclusive nature of available evidence, there is a need for high-quality RCTs assessing their effectiveness and cost-effectiveness. This needs to include trials evaluating the relative effectiveness of alternative ways of delivering interventions. In addition, given the nature of parent-directed interventions, trials should be designed so that the impact of relevant parent, child, and impairment characteristics on effectiveness can be tested. In 2017, Sciberras et al.[57] published a protocol for a large RCT (n=320) that assessed the effectiveness and cost-effectiveness of a comprehensive tailored intervention in improving sleep in children with ADHD. Described as a translational study, it evaluated one of the interventions included in this review[34] in terms of effectiveness (and cost-effectiveness) when delivered in clinical settings by paediatricians or psychologists. Recruitment to this trial was completed in October 2016; however, findings have not yet been published. This RCT will make an important contribution to the evidence base when the results are reported. However, given the diversity of the patient group and the number of non-pharmacological interventions available, additional RCTs and replication studies – conducted in settings where the intervention can be delivered as routine practice and across all (relevant) neurodisabilities – are required. Finally, going forward, we would note the importance of detailed reporting of the interventions using a standardized checklist for describing complex interventions.[58]

Strengths and weaknesses of the research

This review provides a comprehensive overview of the evidence available on non-pharmacological interventions to manage sleep disturbance for children with neurodisabilities. We present only the most commonly reported child sleep outcomes in this paper owing to the vast number of unique outcome measures reported. The full results will be available in an NIHR HTA report (https://www.journalslibrary.nihr.ac.uk/programmes/hta/1421202/#/), which reaches the same conclusions as drawn in this paper. We undertook systematic searches of 16 databases for published, unpublished, and ongoing studies. There were no language restrictions and we included one study published in Chinese. As with all systematic reviews, there was a passage of time between the last date of the literature searches (February 2017) and publication. As a result, there may be one or more studies that have subsequently been published which meet our review's inclusion criteria that are not included here. Our searches of trial registries identified five trials that will be completing over the next couple of years, so an update to this review may then be warranted. Standard methods to reduce error and bias at key stages of the review were used. For example, screening and quality assessment were undertaken independently by two researchers (B.B., C.M., G.S., A.S., A.P.). We developed a typology of parent-directed interventions in the way we believe was most meaningful after discussion among members of the team. We hope this makes a useful conceptual contribution to understanding and specifying such interventions. Although others may have found an alternative way to group these interventions, we do not believe it would change the conclusions of this review. We strictly followed the guidance for completing the Cochrane Risk of Bias tool, meaning that studies were downgraded for lack of blinding, which is difficult to achieve with these types of intervention. This affected one study, which had a low risk of bias on all other criteria.[35] Had we applied the Cochrane criteria 'less strictly', this study would have been rated as having low risk of bias. (It is this intervention that is, as noted earlier, currently subject to a translational trial).[57] This raises an important issue for studies in this area, as classifying studies as having a high risk of bias will mean that non-pharmacological evidence will always seem weaker than studies of pharmacological evidence. At the same time, in the absence of an established method of blinded outcome assessment, there is a risk of overestimating the effectiveness of an intervention where allocation is unblinded and parent-reported outcomes have an element of subjectivity.

Unanswered questions and further research

The substantial health, social, and economic effects of sleep deprivation mean this lack of robust evidence needs to be addressed. A recent UK national research prioritization exercise for children with neurodisability ranked the management of sleep disturbance in the top 10 research priorities.[59] We therefore argue for strategic investment on this topic and our proposed research recommendations are set out below.

However, we note that, on the basis of this review's findings, it is difficult to closely specify where such

research should be focused. We suggest that attention is paid to interventions that are feasible to deliver in routine practice. Furthermore, acknowledging the resource constraints of public services and, as was done by some studies reviewed and where appropriate, evaluations should compare lower and higher intensity modes of delivery (e.g. direct vs remote contact between parent and professional; qualifications of staff delivering the intervention; text-based information/advice vs face-to-face session). Finally, we would argue there is no strong case for developing new interventions. Going forward, the focus should be on further evaluation of existing interventions that appear, on the basis of this review, to have some degree of promise, have (if relevant) been manualized, and are relevant to the ways in which health care is delivered.

First, high-quality RCTs assessing non-pharmacological interventions for sleep disturbance in children with neurodisability are needed. These RCTs should assess the key questions of what works, for whom, and in what circumstances. Intervention development would benefit from being informed by mixed methods research into the mechanisms by which non-pharmacological interventions may affect a child's sleep. A theory-driven approach to intervention development and evaluation is essential if we are to gain understanding of an intervention's active ingredients and the factors that may moderate or mediate their therapeutic action.[60–62]

Second, non-pharmacological interventions for managing sleep disturbance in children with neurodisability are 'complex interventions', made up of several interacting elements. Future research may benefit from adopting the UK Medical Research Council's framework on developing and evaluating complex interventions.[63] This would enable robust approaches to the development and evaluation of complex interventions to be adopted that are grounded in theory.[60] Future research publications should ensure that interventions are described in sufficient detail for replication, for example through use of the Template for Intervention Description and Replication (TIDieR) checklist.[58]

Third, none of the included studies were presented as preventive interventions. The brief, less intense, parent-directed interventions reviewed may align with a preventive or early intervention approach. Evaluating the impact of these interventions on preventing the development of sleep disturbance, or preventing a newly emerging sleep disturbance increasing in severity, would be beneficial.

Fourth, future evaluations should include an economic evaluation, including consideration of costs to families as well as to service providers.

Finally, future research to establish a method of blinded outcome assessment in this area would be beneficial. Additionally, methodologists may wish to consider how to grade lack of blinding in studies where blinding is not possible and outcomes are subjective.

CONCLUSIONS

A wide range of non-pharmacological interventions have been evaluated for managing sleep disturbance in children with neurodisabilities. Although there is some evidence that parent-directed interventions may improve outcomes for children, it was not possible to draw definitive conclusions owing to the lack of robust evidence and substantial heterogeneity across studies. Current clinical guidance recommends parent-directed interventions should be the first approach to managing sleep disturbance; prioritizing research in this area is therefore recommended.

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SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section at the end of the article.

Appendix S1: Search strategies.

Appendix S2: Decision tree for categorizing non-pharmacological interventions.

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