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

ARABELLA SCANTLEBURY1 | CATRIONA MCDAID2 | VICKI DAWSON3 | HEATHER ELPHICK4 | CAROLINE FAIRHURST2 | CATHERINE HEWITT2 | ADWOA PARKER2 | GEMMA SPIERS1 | MEGAN THOMAS5 | KATH WRIGHT6 | BRYONY BERESFORD7

1 Institute for Health and Society, Newcastle University, Newcastle Upon-Tyne; 2 York Trials Unit, Department of Health Sciences, University of York, York; 3 The Children’s Sleep Charity, Balby, Doncaster; 4 Sheffield Children’s NHS Foundation Trust, Sheffield; 5 Blackpool Teaching Hospitals NHS Foundation Trust, Blackpool; 6 Centre for Reviews and Dissemination, University of York, York; 7 Social Policy Research Unit, University of York, UK.

Correspondence to Bryony Beresford, University of York, Social Policy Research Unit, York, YO10 5DD, UK. E-mail: bryony.beresford@york.ac.uk

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 diagnoses9–14 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
and sleep-related movement disorders were excluded. Disorders of hypersomnolence, were included. Central disorders of hypersomnolence on the basis of parental/carer or child report or sleep observations of any duration relating to initiation, maintenance, or scheduling, or quality of 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.

METHOD
The review was conducted in accordance with the Centre for Reviews and Dissemination’s guidance for undertaking reviews in health care and reported according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. The review was prospectively registered with PROSPERO (registration number CRD42016034067). 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. 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 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.

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

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, A Cochrane Risk Of Bias Assessment Tool for Non-Randomized Studies of Interventions for other studies with a control group, or an adapted
checklist for before-and-after studies.\textsuperscript{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.\textsuperscript{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 psycho-educational 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 time-point 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,\textsuperscript{5,26–34} and in all RCTs blinded outcome assessment was either not undertaken or it was unclear whether blinding had occurred.\textsuperscript{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)\textsuperscript{17–50} and likely or unclear bias in measurement of intervention outcomes.\textsuperscript{39,40,43–46,51}

**Outcomes**

Seventy-one sleep-related outcomes were reported across the included studies. Given the number of outcomes...
assessed, in this paper we only report the two most commonly measured outcomes: Child Sleep Habits Questionnaire (CSHQ), 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. 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. 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 and four before-and-after studies 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) | Mean age (SD), y:mo | Neurodisability disorder | Sleep disturbance | Risk of bias |
|-------------------------------------|---------|--------------|-----------------------------------------------|-----------------|---------------|-------------------|-------------------------|------------------|-------------|
| Comprehensive tailored interventions | Austin et al. | 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 | | | | Mixed | Sleep initiation and maintenance | High |
| | Beresford et al. | 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 | | | | Mixed | Sleep initiation and maintenance | High |
| | Beresford et al. intervention 2 | Associated papers | 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 | | | | Mixed | Sleep initiation and maintenance | High |
| | Hiscock et al. | 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 | | | | ADHD: learning disability or ASD/Asperger syndrome | High |
| | Johnson et al. | 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 | | | | Autism and ASD | High |
| | Moss et al. | 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 | | | | Mixed | Sleep initiation, maintenance, and scheduling, and snoring | High |
| | Quine and Wade | 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 | | | | ADHD | Sleep initiation and maintenance | High |
| | Sciberras et al. | 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 | | Mean (SD) not reported (range 3-21y) I: 12:1 (2:2) C: 10:11 (2:6) | | ADHD | Sleep initiation | High |
### Table I: Continued

| Study design | Participants randomized (total n and by group) | Intervention (I) | Comparator (C) | Mean age (SD), y:mo | Neurodisability disorder | Sleep disturbance | Risk of bias |
|--------------|------------------------------------------------|-----------------|----------------|---------------------|--------------------------|------------------|--------------|
| 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.70 | Parallel group RCT n=36 | I: Training curriculum in a booklet given to parent | 6:5 (2:7) (not reported separately) | Mixed | Sleep initiation | High |
| Beresford et al.5 | 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 | 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 |
| Bramble44 | 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 | Parallel group RCT n=80 | I: Training curriculum delivered by two group sessions. Implementation support delivered by telephone calls | I: 5:11 (2.8) | Mixed | Sleep initiation | High |
| Montgomery et al.76 | Parallel group RCT n=82 | Ia: Training curriculum contained in a booklet given to parent | Mean (SD) not reported (range 27–101mo) (not reported separately) | Mixed | Sleep initiation and maintenance | High |
| Reed et al.48 | 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 | 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 | 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) | Mean age (SD), y:mo | Sleep disturbance | Risk of bias |
|--------------------------------------|---------|--------------|-----------------------------------------------|---------------------------------|---------------------|-----------------|-------------|
| Wiggs and Stores<sup>34</sup>        | UK      | Cluster RCT  | n=30                                          | 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)         | Sleep initiation and maintenance | High        |
| Associated papers<sup>73,75</sup>    |         |              |                                              | C: 10:9 (3:10)                  |                     |                 |             |
|                                      |         |              |                                              | Mixed                           |                     |                 |             |
| Other non-pharmacological interventions |         |              |                                              |                                 |                     |                 |             |
| Gringras et al.<sup>32</sup>         | 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) | Sleep initiation and maintenance | High        |
|                                      |         |              |                                              | Mixed                           |                     |                 |             |
| Guilleminault et al.<sup>37</sup>    | 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.<sup>40</sup>            | 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) | Parent/guardian report of sleep dysfunction | High        |
|                                      |         |              |                                              | ASD                             |                     |                 |             |
| Piazza et al.<sup>36</sup>           | USA     | Parallel group RCT | n=14 | 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) | Sleep initiation and maintenance | High        |
|                                      |         |              |                                              | Mixed                           |                     |                 |             |
| Yehuda et al.<sup>46</sup>           | Israel  | Controlled before-and-after study | n=78 | 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 | Sleep deprived | Unclear     |
|                                      |         |              |                                              | ADHD                            |                     |                 |             |
| Yu and Hong<sup>39</sup>             | 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, and two evaluated alternative ways of delivering an intervention: one compared the mode of implementation support (home visit vs telephone call); and the other compared the intensity of practitioner involvement when delivering the intervention (brief vs extended).

CSHQ. Four RCTs (n=310) and two before-and-after studies (n=20) 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). 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). 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).

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

SOL. One RCT (n=40) and two before-and-after studies (n=21) 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) | 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 (clinical) vs two (clinical)                                       | None                                                                                                | 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.59    | 12-16wks                                                         | Face-to-face. Two (clinic, home)                                                    | Fortnightly sessions at clinic Described as ‘weekly’ home visits, although study authors also report frequency decided between practitioner and parent and diminishing in intensity | No                                                | No      | 12wks, 24wks      |
| Quine and Wade43      | 6-28wks                                                          | Face-to-face. Two (home)                                                             | No                                                                                                | 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         |
Randomized controlled trials

| Study            | Baseline, mean (SD) | Follow-up, mean (SD) | Mean difference* (95% CI) |
|------------------|---------------------|----------------------|--------------------------|
| Beresford et al. | I: 59.50 (11.82)   | I: 52.17 (11.44)     | −1.16 (−14.27 to 1.95)*   |
|                  | C: 53.33 (4.27)    | C: 53.33 (8.76)      |                          |
| Hiscock et al.   | I: 57.8 (8.8)      | I: 50.1 (8.3)        | Adjusted: −6.6 (−8.5 to −4.6)* |
|                  | C: 59.0 (7.8)      | C: 55.1 (8.6)        | −5.0 (−7.6 to −2.4)*      |
| Moss et al.      | I: 56.20 (9.38)    | I: 46.50 (7.29)      | −4.62 (−10.83 to 1.59)*   |
|                  | C: 51.38 (7.54)    | C: 51.12 (6.51)      |                          |
| Sciberras et al. | NR                  | (change score) 5.09 (5.12) | −1.73 (−7.11 to 3.65)*   |

Before-and-after studies

| Study            | Baseline, mean (SD) | Follow-up, mean (SD) | Mean difference* (95% CI) |
|------------------|---------------------|----------------------|--------------------------|
| Austin et al.    | 55.43 (7.68)        | 47.57 (9.14)         | −7.86 (−14.39 to −1.33)*  |
| Beresford et al. | 59.55 (7.59)        | 56.57 (10.77)        | Cannot be estimated. Effect size given as 0.42. |

*Unadjusted mean difference unless otherwise stated. bReported in paper. *Difference in change scores from baseline to 2mo between intervention and control groups. 4As not a matched sample (n=11 preintervention and n=7 postintervention). I, intervention; C, comparator; NR, Not reported.

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 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 intervention48 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 −11.248 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 0.02.31

SOL. Two RCTs (n=116)30,31 and two before-and-after studies (n=40)34,46 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 study34,47 evaluated non-comprehensive interventions (Table V).

The RCT (n=30) evaluated an intervention that focused specifically on behavioural principles of managing problem sleep.44 The comparator was an attention control. Neither CSHQ nor SOL were reported in this study. The before-and-after study (n=23) evaluated an intervention47 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 V).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 tapping (mean difference −11.5, 95% CI: −13.3 to −9.7).39
| 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                                                             | Yes                                              | Yes    | 6wks                |
| Before-and-after studies             |                                                                          |                  |                                                   |                                                                        |                                                                          |                                                   |        |                     |
| Beresford et al.42                   | 5wks                                                                     | Group            | Four                                              | Yes                                                                    | None (but included within curriculum for group session)                | No                                               | Yes    | 5wks, 17wks, 29wks  |
| Beresford et al.41                   | intervention 3                                                           | One session      | Teaching workshop                                 | One                                                                    | No                                                                       | No                                               | Yes    | 12wks, 24wks        |
| Beresford et al.41                   | intervention 4                                                           | One session      | Face-to-face (clinic)                             | One                                                                    | Minimal ("only minor individual tailoring")                             | No                                               | Yes    | 2wks, 4mo, 18mo     |
| Bramble44                            |                                                                          | 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    | 7wks                |
| 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.30                          | 7wks                                                                     | Group, plus weekly telephone calls | Three                                            | Yes                                                                    | Weekly for 4wks                                                          | Yes, autism spectrum disorder                      | Yes    | 3wks, 7wks, 11wks   |
| 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\(^{34}\)        | 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 |
| Peppers et al\(^{47}\)           | Principles of sleep hygiene for children with attention-deficit–hyperactivity disorder | One session                    | Face-to-face (clinic) | One (clinic)                  | Yes                                                                                                     | Yes, attention-deficit–hyperactivity disorder                  | Unclear | 6wks                |
| 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  | 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                |
| Gringras et al.32             | 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’ |
| Piazza et al.36               | 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                 |
| Before-and-after studies      | 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          |
| Oriel et al.40                | 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               |
| Yehuda et al.46               |                                               |                                                                                                                                                                                                                       | 2020                                                                                                                       |                                                                   |                     |
SOL. One RCT ($n=73$) and one before-and-after study ($n=8$) measured SOL. There was no statistically significant difference for this outcome in the RCT comparing weighted blankets with placebo blankets (actigraphy-measured SOL: mean difference 2.1, 95% CI: -0.5 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 -19.1, 95% CI: -40.9 to 6.5).

**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 intervention delivered via telephone and mail, rated as being at low risk of bias for all domains except blinding, as being at low risk of bias with ADHD and rated as being at low risk of bias in comparison to a non-comprehensive control group. Two other studies evaluating comprehensive interventions also showed statistically significant improvements in SOL and in parent-reported SOL. One RCT of a comprehensive intervention delivered via a single face-to-face session, one RCT of a comprehensive intervention delivered via telephone and mail, and one before-and-after study of a non-comprehensive intervention (ADHD-specific) also showed statistically significant improvements in parent-reported SOL and in parent-reported SOL. One RCT of a before-and-after study evaluating an aquatic exercise intervention (mean difference 19.1, 95% CI: -0.95 to 6.5).

**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 neurodisability? | 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 |
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., 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.

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. 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 in terms of effectiveness (and cost-effectiveness) when delivered in clinical settings by pediatricians 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.

**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, 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. 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.) 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. 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 RCT’s 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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RESUMEN

INTERVENCIONES NO FARMACOLÓGICAS PARA LA ALTERACIÓN DEL SUEÑO – DE CAUSA NO RESPIRATORIA - EN NIÑOS CON DISCAPACIDADES NEUROLÓGICAS: UNA REVISIÓN SISTEMÁTICA

OBJETIVO Describir la evidencia existente sobre intervenciones no farmacológicas para controlar la alteración del sueño en niños con discapacidades neurológicas.

MÉTODOS Revisión sistemática de intervenciones no farmacológicas destinadas a mejorar la alteración del sueño de causa no respiratoria en niños con discapacidades neurológicas. Se realizaron búsquedas en dieciséis bases de datos, literatura gris y listas de referencias de documentos incluidos hasta febrero de 2017. Dos investigadores llevaron a cabo exámenes de detección, extracción de datos y evaluación de la calidad.

RESULTADOS Se incluyeron veinticinco estudios: 11 ensayos controlados aleatorios (ECA) y 14 estudios observacionales - antes y después. Todos los estudios tenían un riesgo de sesgo alto o poco claro. Las intervenciones dirigidas a los padres se categorizaron como: intervenciones integrales a medida (n = 9), intervenciones integrales no adaptadas (n = 8), intervenciones no integrales (n = 2). Seis “otras” intervenciones no farmacológicas se incluyeron. Se midieron los resultados relacionados con el sueño de 71 niños y padres a través de los estudios incluidos. Presentamos los dos resultados más comúnmente medidos: Cuestionario de Hábitos de Sueño Infantil y Latencia de Inicio del Sueño. Cinco estudios informaron mejoras significativas en al menos uno de estos resultados.

INTERPRETACIÓN Se han evaluado diversos tipos de intervenciones no farmacológicas para controlar la alteración del sueño. Debido a la heterogeneidad clínica y la mala calidad de los estudios no podemos sacar conclusiones definitivas sobre la efectividad de estas intervenciones. La guía clínica actual recomienda las intervenciones dirigidas por los padres como el primer enfoque para manejar la alteración del sueño; se recomienda priorizar la investigación en esta área.

Registro de revisión: registro de PROSPERO: CRD42016034067

RESUMO

INTERVENÇÕES NÃO-FARMACOLÓGICAS PARA DISTÚRBIOS DO SONO NÃO RESPIRATÓRIOS EM CRIANÇAS COM NEURO-INCAPACIDADES: UMA REVISÃO SISTEMÁTICA

OBJETIVO Descrever a evidência existente sobre intervenções não-farmacológicas para manejo de distúrbios do sono em crianças com neuro-incapacidades.

MÉTODOS Revisão sistemática de intervenções não-farmacológicas visando melhorar distúrbios do sono não respiratório em crianças com neuro-incapacidades. Dezesseis bases de dados, literatura cinzenta, e listas de referências dos artigos incluídos foram buscados até Fevereiro de 2017. Dois pesquisadores fizeram a busca, extração de dados, e avaliação de qualidade.

RESULTADOS Vinte e cinco estudos foram incluídos: 11 ensaios clínicos randomizados (ECRs) e 14 estudos antes-depois. Todos os estudos tiveram risco alto ou não esclarecido de viéses. Intervenções direcionadas para os pais foram categorizadas como: intervenções personalizadas compreensivas (n=9), intervenções não personalizadas compreensivas (n=8), intervenções não-compreensivas (n=2). Seis “outras” intervenções não-farmacológicas foram incluídas. Setenta e um desfechos relacionados ao sono de crianças e pais foram mensurados nos estudos incluídos. Relatamos os dois desfechos mais frequentemente mensurados: Questionário dos Hábitos de Sono da Criança, e Latência para Início do Sono. Cinco estudos relataram melhorias significativas em pelo menos uma das medidas.

INTERPRETATION Vários tipos de intervenções não-farmacológicas para manejo dos distúrbios do sono foram avaliados. A heterogeneidade clínica e a pobre qualidade dos estudos implicam que não podemos tirar conclusões definitivas sobre a efetividade destas intervenções. As recomendações clínicas atuais apontam intervenções direcionadas aos pais como a primeira abordagem no manejo de distúrbios do sono; priorizar pesquisas nesta área é recomendado.

Registro da revisão: registro PROSPERO CRD42016034067