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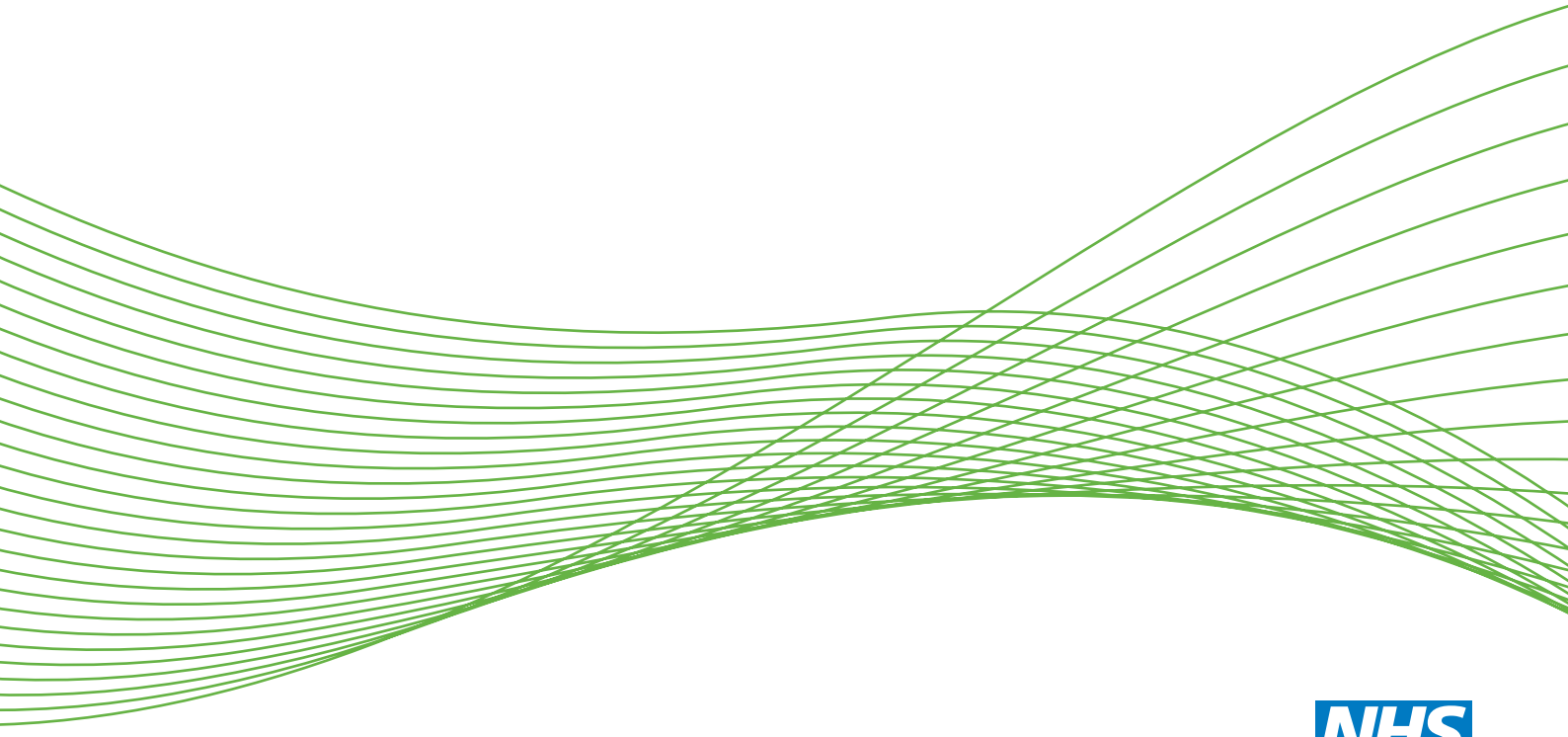

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Pharmacological and non-pharmacological interventions for non-respiratory sleep disturbance in children with neurodisabilities: a systematic review

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Abstract

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

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Background: There is uncertainty about the most appropriate ways to manage non-respiratory sleep disturbances in children with neurodisabilities (NDs).

Objective: To assess the clinical effectiveness and safety of NHS-relevant pharmacological and non-pharmacological interventions to manage sleep disturbance in children and young people with NDs, who have non-respiratory sleep disturbance.

Data sources: Sixteen databases, including The Cochrane Central Register of Controlled Trials, EMBASE and MEDLINE, were searched up to February 2017, and grey literature searches and hand-searches were conducted.

Review methods: For pharmacological interventions, only randomised controlled trials (RCTs) were included. For non-pharmacological interventions, RCTs, non-randomised controlled studies and before-and-after studies were included. Data were extracted and quality assessed by two researchers. Meta-analysis and narrative synthesis were undertaken. Data on parents' and children's experiences of receiving a sleep disturbance intervention were collated into themes and reported narratively.

Results: Thirty-nine studies were included. Sample sizes ranged from 5 to 244 participants. Thirteen RCTs evaluated oral melatonin. Twenty-six studies (12 RCTs and 14 before-and-after studies) evaluated non-pharmacological interventions, including comprehensive parent-directed tailored ($n = 9$) and non-tailored ($n = 8$) interventions, non-comprehensive parent-directed interventions ($n = 2$) and other non-pharmacological interventions ($n = 7$). All but one study were reported as having a high or unclear risk of bias, and studies were generally poorly reported. There was a statistically significant increase in diary-reported total sleep time (TST), which was the most commonly reported outcome for melatonin compared with placebo [pooled mean difference 29.6 minutes, 95% confidence interval (CI) 6.9 to

52.4 minutes; $p = 0.01$]; however, statistical heterogeneity was extremely high (97%). For the single melatonin study that was rated as having a low risk of bias, the mean increase in TST was 13.2 minutes and the lower CI included the possibility of reduced sleep time (95% CI –13.3 to 39.7 minutes). There was mixed evidence about the clinical effectiveness of the non-pharmacological interventions. Sixteen studies included interventions that investigated the feasibility, acceptability and/or parent or clinician views of sleep disturbance interventions. The majority of these studies reported the 'family experience' of non-pharmacological interventions.

Limitations: Planned subgroup analysis was possible in only a small number of melatonin trials.

Conclusions: There is some evidence of benefit for melatonin compared with placebo, but the degree of benefit is uncertain. There are various types of non-pharmacological interventions for managing sleep disturbance; however, clinical and methodological heterogeneity, few RCTs, a lack of standardised outcome measures and risk of bias means that it is not possible to draw conclusions with regard to their effectiveness. Future work should include the development of a core outcome, further evaluation of the clinical effectiveness and cost-effectiveness of pharmacological and non-pharmacological interventions and research exploring the prevention of, and methods for identifying, sleep disturbance. Research mapping current practices and exploring families' understanding of sleep disturbance and their experiences of obtaining help may facilitate service provision development.

Study registration: This study is registered as PROSPERO CRD42016034067.

Funding: The National Institute for Health Research Health Technology Assessment programme.

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Report Supplementary Material 1 Quality assessment checklists

Supplementary material can be found on the NIHR Journals Library report project page (www.journalslibrary.nihr.ac.uk/programmes/hta/1421202/#/documentation).

Supplementary material has been provided by the authors to support the report and any files provided at submission will have been seen by peer reviewers, but not extensively reviewed. Any supplementary material provided at a later stage in the process may not have been peer reviewed.

List of abbreviations

ABC	Aberrant Behavior Checklist
ACROBAT-NRSI	A Cochrane Risk Of Bias Assessment Tool: for Non-Randomized Studies of Interventions
ADHD	attention deficit hyperactivity disorder
ASC	autism spectrum condition
ASD	autism spectrum disorder
ASSIA	Applied Social Sciences Index and Abstracts
BPI	Behavior Problem Index
CBCL	Child Behavior Checklist
CENTRAL	The Cochrane Central Register of Controlled Trials
CI	confidence interval
CSHQ	Children's Sleep Habits Questionnaire
DASS	Depression Anxiety Stress Scales
FISH	Family Inventory of Sleep Habits
HTA	Health Technology Assessment
ICSD-3	*International Classification of Sleep Disorders* – Third Edition
MD	mean difference
ND	neurodisability
PCQ	Parental Concerns Questionnaire
PedsQL	Pediatric Quality of Life Inventory
PSOC	Parenting Sense of Competence
RBS-R	Repetitive Behaviour Scale–Revised
RCT	randomised controlled trial
SD	standard deviation
SE	standard error
SOL	sleep onset latency
TIDieR	Template for Intervention Description and Replication
TST	total sleep time
WASO	wake after sleep onset
WHO	World Health Organization

Plain English summary

Sleep problems such as difficulty settling at bedtime or night waking are more common and serious in children with disorders of development of the brain, for example autism, attention deficit hyperactivity disorder and learning difficulties. Sleep problems can affect children's and parents' mental, physical and emotional well-being, and, so, help with children's sleep is a main concern for parents. There is a wide range of drug and non-drug treatments available to manage sleep disturbance. However, very little is known about whether or not these treatments make a difference; in other words, are these treatments effective?

This study aimed to investigate this gap in knowledge. We reviewed previous research to find out what is already known about the effectiveness of drug and non-drug treatments. Study results suggest that one drug (melatonin) may be helpful for managing children's sleep. However, we cannot tell how beneficial it is. There are also many non-drug treatments (e.g. information leaflets, parent training groups, one-to-one work between a parent and a professional). However, the studies evaluating these sorts of help tested different treatments in different ways. This means that we cannot tell how beneficial each treatment is. A limited number of studies also looked at families' or professionals' views and experiences of these treatments.

Overall, research on treatments of sleep disturbance in children is limited, with only one drug (melatonin) showing signs of benefit. Owing to the limited number of studies available and the largely poor quality of this research, it is not possible to make any suggestions for clinical practice. More research is needed to identify the best treatments for sleep problems among children with neurodisabilities.

Scientific summary

Background

Sleep is essential for physical and mental functioning and well-being. Difficulties with sleep initiation, sleep maintenance and sleep scheduling result in disturbed sleep not only for the individual, but often for other family members as well. Non-respiratory sleep disturbances in children with neurodisabilities (NDs) are more common and more severe than in children with typical development. However, sleep disturbance is rarely a diagnostic criterion; rather, it co-occurs with a diagnosis of a ND. Non-respiratory causes of sleep disturbance among children with NDs include behavioural factors (e.g. parenting), damage or disorders affecting circadian rhythms, hyperarousal, pain, seizures, anxiety and the presence of medical technologies. Many children have more than one sleep difficulty and the aetiology of sleep disturbance is often multifactorial. Sleep disturbances are associated with poor cognitive, physical and/or emotional outcomes for children and parents and result in increased demands on services. Help with sleep is a high priority for parents, practitioners and other stakeholders. However, support is patchy and approaches to managing sleep disturbance are variable and inconsistent. Interventions include pharmacological and non-pharmacological approaches. Pharmacological interventions act on the physiological processes of sleep and/or the timing of the sleep–wake cycle. All are prescribed 'off-label'. Non-pharmacological interventions include parent-directed psychoeducational interventions, chronotherapy, phototherapy, dietary changes and sensory interventions. Evidence on intervention effectiveness is particularly limited for some of these approaches. Importantly, the evidence on pharmacological and non-pharmacological interventions, and across all types of NDs, has never been subject to a single review.

Objectives

- Assess the clinical effectiveness and safety of different intervention approaches to sleep disturbances for children with NDs and, when possible, to:
  - Examine whether or not the clinical effectiveness of an intervention differs for different types of ND, different causes of sleep disturbance and different types of sleep disturbance.
  - Review and evaluate evidence regarding the use of more than one intervention approach, sequentially or in combination, to manage a specific cause of sleep disturbance.
  - Review and evaluate evidence regarding the impact of the setting and/or skills/qualifications of practitioners on intervention effectiveness.
- Assess evidence regarding the acceptability and feasibility of sleep disturbance interventions.
- Describe the settings in which sleep disturbance interventions are being delivered, and by whom.
- Make recommendations, when appropriate, with respect to the management of sleep disturbance among children with NDs generally and/or with respect to particular NDs.
- Identify and describe interventions that look promising and are of relevance and/or feasible to the NHS but that have not been robustly evaluated.
- Make recommendations regarding priorities for future primary research on this topic.

Methods

We undertook a systematic review of pharmacological and non-pharmacological interventions for non-respiratory sleep disturbance among children aged 0–18 years with NDs.

In February and March 2016 (updated in February 2017) we searched Applied Social Sciences Index and Abstracts, The Cochrane Central Register of Controlled Trials, Cochrane Database of Systematic Reviews, Conference Proceedings Citation Index, Cumulative Index to Nursing & Allied Health, Database of Abstracts of Reviews of Effects, EMBASE, Health Management Information Consortium, MEDLINE, MEDLINE In-Process & Other Non-Indexed Citations, PsycINFO, Science Citation Index, Social Care Online, Social Policy & Practice, ClinicalTrials.gov, World Health Organization International Clinical Trials Registry Platform and UK Clinical Trials Gateway. The Social Care Online, Social Policy & Practice, Health Management Information Consortium, Conference Proceedings Citation Index and PsycINFO all provide some coverage of reports and other unpublished documents, so the available grey literature are represented in the search results. The reference lists of relevant systematic reviews and included studies were also scanned. There were no limits on date, language or study designs. Studies of children and young people with NDs experiencing non-respiratory sleep disturbances were included. Studies of NHS-relevant pharmacological and non-pharmacological interventions targeted at improving sleep in any setting with and without a comparator were eligible. For pharmacological interventions, only randomised controlled trials (RCTs) were included. For non-pharmacological interventions, RCTs and other study designs, with and without a comparator, were eligible. Qualitative and quantitative studies of parents' or children's experiences of receiving a sleep disturbance intervention were included. Key study characteristics and results were extracted and quality assessed independently by two researchers. When possible, a meta-analysis was undertaken and, when feasible, subgroup analyses considering previous interventions and NDs were undertaken. When meta-analysis was not possible, a narrative synthesis approach was adopted. Qualitative and quantitative data on parents' or children's experiences of receiving a sleep disturbance intervention were collated into themes or topic areas. A descriptive narrative of these findings was produced.

Results

A total of 39 studies were identified: 25 RCTs and 14 before-and-after studies (one with a control group). Sample sizes varied (range 5–244 participants). Thirteen RCTs investigated a pharmacological intervention, namely the use of oral melatonin. Twenty-six studies investigated non-pharmacological interventions. Nine of these evaluated parent-directed tailored interventions, eight evaluated parent-directed non-tailored interventions and two evaluated non-comprehensive parent-directed interventions. A further seven studies evaluated other non-pharmacological interventions: dietary interventions, $n = 2$; alternative medicine, $n = 1$; exercise-based interventions; $n = 1$; weighted blankets, $n = 1$; faded bedtime with response costs, $n = 1$; and light therapy with a daytime activities programme, $n = 1$.

With the exception of one study, all studies were rated as having a risk of bias. Findings from the pharmacological interventions suggest that there was evidence of benefit with melatonin compared with placebo. There was a statistically significant increase in diary-reported total sleep time (TST), which was the most commonly reported outcome, with melatonin compared with placebo [pooled mean difference 29.6 minutes, 95% confidence interval (CI) 6.9 to 52.4 minutes; $p = 0.01$]; however, the statistical heterogeneity was extremely high (97%). For the single melatonin study that was rated as having a low risk of bias, the mean increase in TST was 13.2 minutes and the lower CI included the possibility of reduced sleep time (95% CI –13.3 to 39.7 minutes). It is difficult to draw conclusions as regards the clinical effectiveness of the non-pharmacological interventions, owing to the limited number of RCTs, the variation and range of outcome measures and the risk of studies being underpowered to detect an effect.

Sixteen of the interventions included in the clinical effectiveness review were also investigated in terms of their feasibility and/or acceptability and/or the parent/clinician views of the intervention. These outcomes are referred to as 'family experience' data; however, such data were limited. The majority of studies used quantitative methods to investigate family experiences of non-pharmacological interventions with the exception of one study, which reported on difficulties with administering medication.

Conclusions

The evidence on the management of sleep disturbances in children with NDs is limited and largely of poor quality. There is some evidence of benefit for melatonin compared with placebo. The extent of this benefit is uncertain and so it is not possible to make recommendations for practice. There is a range of non-pharmacological interventions for sleep disturbance. The clinical effectiveness of these interventions is unclear, owing to the limited number of RCTs, the heterogeneous nature of the interventions and outcomes used and the insufficient power in studies to detect any effect.

Implications for health care

It has not been possible to draw conclusions about the clinical effectiveness of pharmacological or non-pharmacological interventions owing to the quality of research and the lack of available evidence.

Recommendations for research

- The development of a core set of outcome measures would facilitate the evaluation of future assessments of the impact of pharmacological and non-pharmacological interventions for sleep disturbance.
- Further exploration of existing tools, practices and strategies to identify sleep problems in children with ND disorders in routine practice would be beneficial.
- Trials comparing slow-release and fast-release melatonin may be useful.
- Further investigation of combined or sequential use of melatonin (or other pharmacological interventions) and parent-directed interventions is suggested.
- Trials evaluating parent-directed interventions are needed. They should include an exploration of intervention feasibility and acceptability. Interventions addressing sleep initiation should be prioritised.
- Evaluations of low-intensity parent-directed interventions – evaluated to date in children with recognised sleep disturbance – as preventative interventions are recommended.
- Research that maps current practices and explores families' understanding of sleep disturbance, and their experiences of obtaining help, is suggested in order to inform developments in service provision.
- All studies should seek to include a health economics evaluation.

Study registration

This study is registered as PROSPERO CRD42016034067.

Funding

Funding for this study was provided by the Health Technology Assessment programme of the National Institute for Health Research.

Chapter 1 Background

Introduction

This project was undertaken in response to a commissioning brief from the National Institute for Health Research (NIHR) Health Technology Assessment (HTA) programme. The call was for a systematic review to address the question of which interventions – pharmacological and non-pharmacological – are clinically effective for non-respiratory sleep disturbances in children with neurodisabilities (NDs) and which have generalisable, as opposed to disorder-specific, effects. The brief requested a broad systematic review to 'take stock' of the current evidence that is available on what works and for whom and to identify promising interventions for future primary research. The following two sections describe the definitions used to identify the scope of the review; we then move on to discuss, specifically, the issue of sleep disturbance in children with NDs.

Sleep disturbance

Sleep has been described as an active 'restorative process'[1] and is essential for optimal physical and mental functioning and well-being. It is a complex process: the timing, duration and quality of sleep is the outcome of the interplay of biological processes, socioenvironmental influences and behaviours. As a result, there is a wide range of reasons why an individual's sleep may be affected in some way and, for children with NDs, the cause may be multifactorial.[2–9]

The *International Classification of Sleep Disorders* – Third Edition (ICSD-3)[10] lists the current diagnostic categorisation of sleep disorders as follows: insomnia, sleep-related breathing disorders, central disorders of hypersomnolence (e.g. narcolepsy), circadian rhythm sleep–wake disorders, parasomnias, sleep-related movement disorders and 'other sleep disorders'.

The extent to which sleep is disturbed, or interrupted, is a diagnostic criterion for insomnias and circadian rhythm sleep–wake disorders. That said, it is accepted that there needs to be room for clinical judgement regarding the clinical significance of the extent of sleep disturbance.[11]

However, although the ICSD-3 provides a classification and diagnostic framework, it is important to note that, in paediatric research at least, very few studies actually use the ICSD-3 criteria to define or screen research participants.[12,13] Instead, in both the clinical and research literature, a number of different phrases are used to describe the manifestations of a sleep disorder in terms of the impact that it has on an individual's sleep: sleep disturbance, sleep problems and sleep difficulties.[2] Such terms have all been used for issues related to falling asleep (i.e. sleep initiation) and staying asleep, as opposed to night wakings or very early waking (i.e. sleep maintenance). The scope of this review was guided by the commissioning brief, which made two clear specifications: it should be concerned with 'non-respiratory sleep disturbance' and sleep disturbance (experienced by the child and/or parent) should be a feature of the presenting problem. The ICSD-3 classification was use to specify sleep disorders that were relevant, or not, to the review. Three types of sleep disorder were not relevant because disturbed sleep is not a diagnostic feature, namely sleep-related breathing disorders, central disorders of hypersomnolence (e.g. narcolepsy) and sleep-related movement disorders. Interventions that addressed sleep disturbances that aligned with the diagnostic features of insomnias or circadian sleep–wake cycle disorders were included. In addition, we included parasomnias because of the potential impact that they could have on parental sleep. However, for reasons noted above, we did not require studies to use the ICSD-3 to define or screen participants. We briefly set out the ICSD-3 definitions of these disorders in the following sections.

Insomnia disorders

The ICSD-3 definition of insomnia is as follows: a persistent difficulty with sleep initiation, duration, consolidation or quality, which occurs despite adequate opportunity and circumstances for sleep and results in some form of daytime impairment. It comprises three subcategories: (1) chronic insomnia disorder, (2) short-term insomnia disorder and (3) other insomnia disorders. In children, chronic insomnia also includes behavioural insomnia disorders, the aetiology of which is located in the practices of parents around bedtime/settling and responding to night/early-morning wakings.[12] Behavioural insomnias are common in childhood, with an even higher prevalence among children with NDs.[14]

Circadian rhythm sleep–wake disorders

Circadian rhythm sleep–wake disorders are characterised by abnormalities in the length, timing and/or regularity of the sleep–wake cycle relative to the day–night cycle. It is caused by genetic, neurological or visual pathway damage/disorders affecting circadian rhythms, including melatonin release.[14,15]

Parasomnias

Parasomnias were included in the review because of the impact that they have on parents' sleep. The ICSD-3 defines parasomnias as sleep-related occurrences that represent undesirable physical or cognitive experiences (e.g. sleep terrors, sleep walking) occurring out of sleep, during the transition from sleep to the awake state or from the awake state to sleep. They are more common in children than in adults.[16]

Childhood neurodisability

A consensus definition offered by Morris *et al.*[17] defines NDs as:

> *. . . congenital or acquired long-term conditions that are attributed to impairment of the brain and/or neuromuscular system and create functional limitations.*

A wide range of conditions fall under this definition, including cerebral palsy, autism spectrum disorder (ASD), Down syndrome and other chromosome disorders, epilepsy, attention deficit hyperactivity disorder (ADHD), neurometabolic degenerative conditions (e.g. Batten disease), genetic disorders (e.g. Rett syndrome), as well as non-specific diagnoses such as 'learning/intellectual disability' and 'developmental delay'. The involvement of the brain and/or neuromuscular system means that children can experience a range of impairments (e.g. sensory, learning, physical/motor function and speech and language) and health complications or clinical needs (e.g. respiratory, orthopaedic, gastroenterological and pain management). The severity of impairment can range from mild to profound. The fact that it is not uncommon for some conditions to co-occur adds to the complexity and severity of impairment. It is not surprising, therefore, that many children with NDs are frequent users of the health service at all levels: community, primary care inpatient and outpatient settings.[18]

In terms of epidemiology, some NDs are quite common (e.g. autism affects ≈1 in 100 children, cerebral palsies affect ≈1 in 400 children and severe intellectual disabilities affect ≈3 in 1000 children). However, also within this 'cluster' of conditions are very rare syndromes [e.g. tuberous sclerosis (incidence of < 1 in 100,000) and ataxia telangiectasia (incidence of < 1 in 40,000)].[19] Estimates of the overall prevalence of ND among the child population in England vary depending on the measure/indicator used and all are flawed in some way. However, it is generally accepted that ≈4 in 100 children have a ND and that children with NDs constitute the largest group of disabled children.[19]

Sleep disturbances in children with neurodisabilities

Sleep disturbances in children with NDs are more common and more severe than in children with typical development.[14,15] However, non-respiratory sleep disturbance is very rarely a diagnostic criterion. Indeed, this is the case only with respect to four conditions: Rett syndrome, Angelman syndrome, Smith–Magenis syndrome and Williams syndrome.[20]

The impacts of sleep disturbance

Sleep disturbance can have an impact on all members of the family: the child and their parents and siblings. Indeed, often it is the parents' own sleep deprivation or poor sleep quality that precipitates them to seek help with their child's sleep.[9,21] Child sleep problems are associated with poor outcomes for parents, such as heightened levels of parental stress and irritability,[22,23] and for children, including poorer educational progress and daytime behaviour problems.[24] These outcomes in themselves increase demands on statutory services, as well as creating further additional support needs, such as respite care.[25,26] The wider association between sleep quality and economic consequences has also been described.[27,28] Parents consistently highlight the need for support with their child's sleep problems,[29,30] although, historically, little time has been allocated to training the relevant professionals to provide this kind of support.[31] Parents, practitioners and other stakeholders agree that research on sleep management interventions is a priority with respect to children with NDs.[32]

Interventions for non-respiratory sleep disturbance

Interventions to address sleep disturbance among children with NDs examine both pharmacological and non-pharmacological approaches.

Pharmacological interventions act on an aspect of the physiological processes of sleep and/or the timing of the sleep–wake cycle; the most frequently used interventions are melatonin (a hormone playing a key role in the timing of the sleep–wake cycle), clonidine (which inhibits noradrenaline activity and hence has a soporific effect) and antihistamines [which inhibit neurotransmitters (histamines) that are involved in wakefulness/alertness].[33–35] All are prescribed 'off-label'. Other pharmacological interventions are used in relation to children's sleep, such as medications to manage seizures and pain; however, these are outside the scope of this review because the primary purpose of the intervention is not sleep disturbance.

Non-pharmacological interventions address other causes of disturbances in sleep initiation, maintenance and/or scheduling and are wide-ranging in approach. Interventions that are available within and/or outside the NHS include:

- behavioural and cognitive behavioural interventions – addressing behavioural aspects of sleep including parents' management of sleep behaviours and routines
- chronotherapy – intervening in the timing of sleep within the 24-hour cycle
- phototherapy (or 'bright light therapy') – using light exposure to effect changes in the circadian rhythm
- dietary interventions – removing stimulants and restricting to hypoallergenic food
- sensory interventions, including weighted blankets[36] and 'safe space' bed tents[37]
- cranial osteopathy[38]
- changing the bedroom environment, for example by removing any televisions or other stimulatory materials and adjusting heating and/or lighting.

Current guidance on the management of sleep disturbance in children advocates that once clinical (e.g. pain or seizures) or respiratory reasons for sleep disturbance are excluded, behavioural approaches that seek to change parents' responses to sleep-related problems should be the 'first port of call' for any child,[33,39–41] with pharmacological intervention (and to date, this is typically melatonin) suggested in cases in which such approaches prove ineffective, which should be used alongside behavioural parent-directed approaches.[2,42]

The justification for this is that, in common with children with typical development, the origins of sleep difficulties for many disabled children are behavioural, located in the way in which parents address and manage their child's sleep.[4,43,44] The intensity of behavioural intervention depends on the complexity of the sleep problem and/or child/family-centred factors. Sleep problems that cannot be resolved through low-intensity approaches (e.g. simple information leaflets, verbal information/guidance during a routine appointment, one-off 'sleep management workshops') may require a more tailored and sustained approach involving a detailed assessment of the sleep problem, the creation of a bespoke sleep management strategy (based on behaviour modification principles) and time-limited (typically face-to-face) support as parents implement the strategy. In recent years, there has also been an increasing interest in using groups, as opposed to a series of one-to-one sessions, to deliver behavioural sleep support to parents.[45] Furthermore, online 'self-directed' sleep management interventions for adults are now available (e.g. Sleepio.com), and the management of sleep disturbance also appears within the curricula of newly available online parenting interventions for parents of children with typical development and disabilities (e.g. Stepping Stones Triple P). Constrained resources and wider changes in the way in which health care is delivered mean that it is likely that these newer modes of delivering behavioural sleep management intervention, such as groups and self-management, will increasingly be considered and used by clinicians and parents.[46] It was important, therefore, that this review examined evidence of the impact that the mode of delivery has on outcomes.

The availability of sleep interventions and the organisation of sleep disturbance services

A number of different services deliver a sleep management intervention to children with NDs. They include community paediatric teams, general practitioners, health visitors, specialist paediatric neurology/autism/ADHD services, child and adolescent mental health services and tertiary sleep services. Within the UK, third-sector organisations (e.g. Sleep Scotland, Scope, the Children's Sleep Charity) are also highly active in this area. Such organisations offer education/training to parents and professionals on sleep and behavioural approaches to managing sleep difficulties, as well as sleep intervention services. Some NHS trusts are commissioning services of this kind from third-sector organisations such as these.

Although there has been no systematic analysis of the way that sleep disturbance in children with NDs is managed by the NHS, a recent survey of paediatricians by the British Paediatric Respiratory Society led to the conclusion that services for sleep disorders in children were 'chaotic and unplanned . . . often unfunded and frequently perceived as inadequate for local needs'.[47] Current practice in prescribing medicines such as melatonin has been described as haphazard[48] and access to behavioural interventions as patchy.[49]

There are a number of reasons for this current state of affairs, including the apparent absence of education on sleep disorders and sleep management in medical school education,[50] a lack of recognition of the importance of assessing/checking for sleep disturbance, lack of knowledge/skills or resources to deliver non-pharmacological interventions, parental expectations regarding their child's sleep, the complexity and range of conditions falling under the umbrella of NDs and the complexity of sleep disturbance and its potential causes. In addition, although there have been some attempts to develop sleep management pathways within paediatrics, these have been restricted to particular types of sleep disturbance and/or sleep intervention and/or diagnostic groups, for which the evidence is more plentiful and/or of higher quality.[47,51,52]

Informing the development of a robust evidence base

A robust evidence base is clearly required to inform the development of a paediatric ND sleep management pathway for non-respiratory disturbance that integrates pharmacological and non-pharmacological interventions. A first step towards this, as noted in the commissioning brief, is to 'take stock' of the existing evidence base. Our preliminary scoping found that previous systematic reviews have mainly focused on either individual NDs[51,53–57] and/or single interventions or pharmacological interventions only.[7,53,56,57]

Three reviews that included pharmacological and non-pharmacological interventions were restricted by type of population: children with ASDs,[51] children aged 0–12 years with cerebral palsy or traumatic brain injury[58] and children with ADHD.[54] One review with a wide population of chronic health conditions in patients aged up to 19 years (including NDs) investigated non-pharmacological interventions (behavioural and non-behavioural);[1] another review of behavioural interventions focused on disabled children aged up to 8 years.[59] The search end dates for the previous reviews we identified ranged from 2004 to 2013. None of the previous reviews addressed the research question in the commissioning brief and a new review was considered appropriate.

Given the complexity of sleep disturbance in children with ND, and in contrast to previous reviews, it was essential that our review evaluated pharmacological *and* non-pharmacological interventions, as much as existing evidence allows, across the range of NDs and age groups. This allows identification of evidence gaps, as well as the accumulation of knowledge on which sleep disturbance interventions work (solely or in combination with other interventions) and for whom they work within the diverse population of children with NDs.

Aims and objectives

There were two overarching aims of this review: (1) to identify the implications for practice for non-respiratory sleep disturbance in children with NDs, evidence permitting, and (2) to inform the focus and priorities of a future call by NIHR for primary research in this area. Unlike previous systematic reviews, we have sought to be holistic in terms of both the population (all children with a ND) and the types of intervention (i.e. pharmacological and non-pharmacological). The objectives were to:

- evaluate and compare the clinical effectiveness of different intervention approaches to sleep disturbances for children with NDs and, when possible, to:
    - examine whether or not intervention clinical effectiveness differs for different types of ND, different causes of sleep disturbance and different types of sleep disturbance (i.e. sleep initiation, sleep maintenance and sleep scheduling)
    - review and evaluate evidence regarding the use of more than one intervention approach, sequentially or in combination, to manage a specific cause of sleep disturbance
    - review and evaluate evidence regarding the impact that the setting and/or skills/qualifications of practitioners have on intervention clinical effectiveness
- describe and compare evidence regarding the acceptability and feasibility of sleep disturbance interventions
- describe the settings in which sleep disturbance interventions are being delivered, and by whom
- make recommendations, when appropriate, with respect to the management of sleep disturbance among children with ND generally and/or with respect to particular NDs
- identify and describe interventions that look promising and are of relevance to and/or feasible for the NHS but that have not been robustly evaluated
- make recommendations regarding priorities for future primary research on this topic
- disseminate the findings in a timely and effective way.

Chapter 2 Methods

This systematic review was undertaken in accordance with *Systematic Reviews: CRD's Guidance for Undertaking Reviews in Health Care*[60] and reported in line with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses.[61]

The review protocol was published prospectively and was registered with PROSPERO as CRD42016034067 (see www.crd.york.ac.uk/PROSPERO/display_record.php?RecordID=34067??).

Inclusion and exclusion criteria

Studies were assessed for eligibility based on the criteria detailed in the following sections.

Population

Studies of children and young people with NDs who were experiencing non-respiratory sleep disturbances were eligible for inclusion in the review.

- Children and young people aged from 0 to 18 years were eligible. We did not expect to find many studies targeted at very young infants. Some previous reviews have used a lower age cut-off point of 3 months and others have not. Given the comprehensive nature of the review, we did not use a lower age cut-off point. ND was defined in accordance with the consensus definition developed by Morris *et al.*:[17]

  > *. . . congenital or acquired long-term conditions that are attributed to impairment of the brain and/or neuromuscular system and create functional limitations.*

- Non-respiratory sleep disturbances, of any duration, related to the initiation, maintenance or scheduling of sleep, diagnosed by a health-care professional based on parental/carer or child report or sleep observation were eligible.
- Excluded non-respiratory sleep disorders were central disorders of hypersomnolence (in which daytime sleepiness is not caused by nocturnal sleep disturbance or misaligned circadian rhythms) and sleep-related movement disorders.

We excluded studies of respiratory-related sleep disturbances. However, NDs are complex conditions and sleep disturbances may have multifactorial causes. Therefore, we included studies in which the respiratory-related component was being controlled and the focus of the intervention was another cause of sleep disturbance. We also excluded studies in which the main focus of the intervention was not treatment of the sleep disturbance (e.g. interventions to control seizures when sleep outcomes were also reported) and studies of mixed populations of children with and without a ND, unless the results were reported separately for the two groups or the sample was predominantly (> 90%) children with a ND.

Intervention

NHS-relevant pharmacological and non-pharmacological interventions targeted at improving sleep initiation, maintenance, scheduling or sleep quality in any setting were eligible for inclusion. For pharmacological studies, 'NHS relevant' was defined as relating to drugs that are licensed for use for this indication in children or that are currently used for this purpose in the NHS. For non-pharmacological studies, 'NHS relevant' was defined as those interventions meeting current practice standards; for example, behavioural interventions that used punishment were excluded. Multicomponent interventions were eligible.

The NHS-relevant pharmacological interventions were melatonin, clonidine and antihistamines.

The NHS-relevant non-pharmacological interventions included (but were not restricted to):

- behavioural interventions delivered in a range of settings such as primary, secondary and tertiary or community, outpatient or inpatient that were delivered in groups or to individual children/families by health-care professionals
- self-help booklets, web-based packages and other online support
- behavioural/cognitive–behavioural interventions addressing behavioural aspects of sleep, including parents' management of sleep behaviours and routines
- chronotherapy – intervening in the timing of sleep within the 24-hour cycle
- phototherapy (or 'bright light therapy') – using light exposure to effect changes in the circadian rhythm
- dietary interventions – removing stimulants, restricting to hypoallergenic food
- sensory interventions, including weighted blankets and 'safe space' bed tents
- cranial osteopathy
- changing the bedroom environment, for example by removing any televisions or other stimulatory materials and adjusting heating and/or lighting.

Comparator

Studies using no intervention, waiting list control, placebo or another NHS-relevant intervention were eligible for inclusion.

Outcomes

The following outcomes were assessed.

Primary outcomes:

- child's sleep-related outcomes – parent-/carer- and child-reported outcomes relating to the initiation, maintenance, scheduling or quality of sleep (using measures such as sleep diaries, standardised scales, e.g. the Composite Sleep Disturbance Index or Epworth Sleepiness Scale) and objective measures such as actigraphy (used to calculate outcomes such as total sleep duration, time taken to fall asleep or sleep efficiency)
- parent sleep-related outcomes – quality of sleep
- measures of perceived parenting confidence and/or efficacy and/or understanding of sleep/sleep management (which are particularly relevant for parent training/behavioural interventions that seek to change the way that parents manage sleep disturbance).

Secondary outcomes:

- child-related quality of life, daytime behaviour and cognition
- parent/carer quality of life and well-being, including global quality of life (e.g. Short Form questionnaire – 36 items) and more specific outcomes such as physical well-being, mental well-being, and mental health (e.g. stress, depression)
- family functioning
- adverse events, including side effects from medication.

Data on uptake of the intervention, retention and intervention adherence were used as indicators of the acceptability and feasibility of the intervention. Quantitative or qualitative data on parents'/children's experiences of receiving a sleep disturbance intervention included:

- the acceptability and feasibility of the intervention
- other experiences of receiving the intervention
- satisfaction with intervention outcomes and 'fit' with their priorities with regard to their child's sleep disturbances; views/perspectives on the mechanisms by which outcomes were achieved.

Study design

Randomised controlled trials (RCTs) and non-randomised controlled studies, such as controlled before-and-after studies and cohort studies with a control group, were included. Both parallel and crossover RCTs were eligible for inclusion. Concerns have been expressed by others that a crossover design may be inappropriate owing to uncertainty about the duration of the effect of interventions on sleep patterns and circadian rhythm and, therefore, on the most appropriate duration for the washout period.[48] We agree with these concerns. However, given that the aim was to undertake a broad review and, as there were few RCTs likely to be available, we included crossover studies.

In order to achieve the second objective of the review, studies without a control group were included in the absence of controlled studies, that is, cohort studies and before-and-after studies. This was because they could include potentially promising interventions that were at an early stage of evaluation. Case studies were not eligible for inclusion.

Qualitative and quantitative studies were included if they reported data on parents' or children's experiences of receiving a sleep disturbance intervention (including intervention acceptability), such as the process of receiving the sleep intervention, satisfaction with intervention outcomes and 'fit' with their priorities with regard to their child's sleep disturbances, and views/perspectives on the mechanisms by which outcomes were achieved. Data could be collected as part of studies of clinical effectiveness or studies that only sought to examine research questions on experiences and satisfaction.

Literature searches

The available evidence was identified by carrying out systematic searches of electronic databases, and reference checking of relevant reviews and included studies. The list of included studies identified from the electronic searches was shared with clinicians in the team to establish if there were any relevant studies that were missing. The searches were undertaken by an experienced information specialist and the search strategy was peer reviewed by a second information specialist.

Information sources

A range of databases were searched in February and March 2016 and updated in February 2017 to ensure coverage from the fields of health, nursing and allied health, and social care. We searched Applied Social Sciences Index and Abstracts (ASSIA), The Cochrane Central Register of Controlled Trials (CENTRAL), Cochrane Database of Systematic Reviews, Conference Proceedings Citation Index, Cumulative Index to Nursing & Allied Health, Database of Abstracts of Reviews of Effects, EMBASE, Health Management Information Consortium, MEDLINE, MEDLINE In-Process & Other Non-indexed Citations, PsycINFO, Science Citation Index, Social Care Online and Social Policy & Practice.

The Social Care Online, Social Policy & Practice, Health Management Information Consortium, Conference Proceedings Citation Index and PsycINFO all provide some coverage of reports and other unpublished documents; therefore, the available grey literature are represented in the search results.

In addition, ClinicalTrials.gov, the World Health Organization (WHO) International Clinical Trials Registry Platform and the UK Clinical Trials Gateway were searched for trials, both ongoing and completed. No limits on date, language or study design were applied in the searches. Full details of search strategies used and numbers of records retrieved are given for each database in *Appendix 1*.

While the search results were being scanned, some additional search terms that had not been included in the original search strategies were identified. Consequently, we carried out some further searches of each of the databases that incorporated these new terms alongside the original search strategy. Details of these additional search strategies can be found in *Appendix 2*.

The reference lists of relevant systematic reviews and included studies were also scanned.

Screening and study selection

The database search results were all loaded into EndNote bibliographic software [version 17.0.2.7390, Clarivate Analytics (formerly Thomson Reuters), Philadelphia, PA, USA] and deduplicated.

There was a three-stage screening process to manage the large number of records. First, the titles of the records were screened for relevance. Two researchers did this jointly for 10% of the titles, and the remainder were screened independently by a single researcher. Records that were identified as potentially relevant based on their title were screened independently by two researchers. When there was no consensus, a third member of the team was consulted. The full texts of potentially relevant papers were ordered. Finally, full papers were independently screened against the eligibility criteria by two researchers. Disagreements were resolved by consensus or by consultation with a third team member if necessary.

Data extraction

A data extraction form for study details was developed and piloted. A Microsoft Excel 2010® spreadsheet (Microsoft Corporation, Redmond, WA, USA) was used to extract the outcome data. Owing to the various ways in which adverse events were described in papers, data for this outcome were extracted separately into a table using Microsoft Word 2010® (Microsoft Corporation, Redmond, WA, USA). All data were extracted by one researcher and checked by a second. Details of the data items extracted are available in *Appendix 3*. For the purposes of this review, we extracted follow-up data relating to the assessment time point closest to the end of the intervention.

Assessment of risk of bias

The Cochrane risk of bias tool[62] was used to assess the quality of RCTs and the newly developed tool, A Cochrane Risk Of Bias Assessment Tool: for Non-Randomized Studies of Interventions (ACROBAT-NRSI), was used to assess the non-randomised controlled before-and-after studies.[63] Uncontrolled before-and-after studies were assessed using questions adapted from ACROBAT-NRSI, as used in another HTA review.[64] Risk of bias was independently assessed by two researchers. Disagreements were resolved through consensus and through discussion with a third researcher if necessary. In addition, for crossover trials, we assessed whether or not an appropriate analysis using paired data was conducted and whether or not there was a treatment by period interaction, as undertaken in a previous systematic review including crossover studies.[65]

A summary risk-of-bias score was calculated following guidance.[62] This score was calculated as follows: any study that had one or more of the domains on the risk-of-bias tool classified as ‘no’ was considered to be rated as having a high risk of bias. Any study that had one or more domains classified as ‘unclear’ on the risk-of-bias tool was considered to be rated as having an unclear risk of bias. To be considered to have a rating of a low risk of bias, a study needed to meet the criteria on all domains on the risk-of-bias tool and classify them as ‘yes’.

For studies containing qualitative and quantitative data on parents' and/or children's satisfaction with the intervention, take-up, retention and adherence to the intervention and experiences of the intervention, the quality of the study was assessed and reported using the quality appraisal checklist of Hawker *et al.*[66]

It was not possible to blind the types of non-pharmacological interventions and comparators used in the studies under consideration. In addition, owing to the nature of the outcomes measured, robust, blinded outcome assessment was difficult. Although actigraphy-based child sleep outcomes are more objective than parent-reported measures, we did not consider these to be true objective outcomes, with non-blinding unlikely to introduce bias. Therefore, all of the measures were regarded as having the capacity to be influenced by lack of blinding.

Analysis

The synthesis aimed to:

1. Assess the clinical effectiveness of the interventions for sleep disturbance, in particular interventions that may work across conditions.
2. Inform future research by identifying gaps in the evidence and identifying interventions that are the most promising front runners to be considered for future primary research.

First, narrative and tabular summaries of key study characteristics were undertaken. This allowed a mapping of which interventions have been investigated for which ND and for which type of sleep disturbance (e.g. sleep initiation) in order to identify interventions that have been investigated across conditions. We also mapped information on the feasibility and acceptability of each of the interventions.

Synthesis involved paired meta-analyses and narrative synthesis.

Meta-analyses

Pharmacological intervention studies

When sufficient data for our primary and secondary outcomes were available, they were pooled in quantitative synthesis using a random-effects model (for continuous outcomes). As data sets often included both parallel and crossover trials, or just crossover trials, data were pooled using the generic inverse variance method in RevMan version 5 (The Cochrane Collaboration, The Nordic Cochrane Centre, Copenhagen, Denmark).[67] Only crossover trials with a washout period were included in meta-analyses, for which data from both treatment periods were used. For trials without a washout period, we considered using data from the first period only; however, in the event, this was not possible, as data summaries were not provided by treatment period for these trials.

The recommendations provided in the Cochrane handbook were followed as closely as possible.[68] The mean difference (MD) between melatonin (M) and placebo (P) at the end point was either taken as reported in the article or calculated as the difference in means for each group/period:

$$\text{MD} = \text{mean}_{\text{M}} - \text{mean}_{\text{P}}. \quad (1)$$

When a 95% confidence interval (CI) for the group means was presented instead of a standard deviation (SD) (e.g. Garstang and Wallis[69]), the SD was calculated using the formula:

$$\text{SD} = \sqrt{N} \times (\text{upper confidence limit} - \text{lower confidence limit})/(2 \times t - \text{value}), \quad (2)$$

where the *t*-value was obtained by entering = tinv(1–0.95, N-1) in a cell in a Microsoft Excel spreadsheet.

For the crossover trials, when only means and SDs for the measurements of the intervention (melatonin) and the control (placebo) were available, the SD of within-participant differences between M and P measurements was estimated using the formula:

$$SD_{diff} = \sqrt{SD_M^2 + SD_P^2 - (2\rho SD_M SD_P)}. \quad (3)$$

The correlation coefficient ρ was estimated from other studies reporting all three SDs for the same outcome or by using 0.5 when no other studies were available to use.

The standard error (SE) of the MD was calculated using:

$$SE\ (MD) = SD_{diff}/\sqrt{N}, \quad (4)$$

where N is the sample size, or by dividing the MD by the t-statistic when this was presented for a two-sample t-test of the period differences (e.g. Weiss *et al.*[70]).

To transform the parallel-group trial data for entry into the generic inverse variance facility, a two-sample t-test was conducted to calculate the unadjusted difference (and SE of the MD) between the groups at follow-up (post intervention) using raw group summary data (N, mean and SD).

Statistical heterogeneity between trials was assessed using the I^2 statistic.[71] Two sources of potential clinical and methodological heterogeneity were identified for the pharmacological intervention trials, and subgroup analyses were conducted based on these, when appropriate:

1. type of neurological disorder – population primarily with ASD
2. receipt of prior intervention – whether or not participants were offered an additional intervention prior to the start of the study.

The risk of publication bias was not formally assessed.

When data could not be pooled, summaries of the findings for each trial and outcome are presented with a (estimated unadjusted) MD and 95% CI between melatonin and control at follow-up.

Adverse event data are summarised narratively.

Non-pharmacological intervention studies

Non-pharmacological intervention studies included parallel-group RCTs and before-and-after studies. Owing to insufficient data for each outcome and/or significant heterogeneity in study design and intervention, data were not pooled in meta-analyses. Narrative and quantitative summaries of the findings for each trial and outcome are presented. For continuous data in the RCTs, the preferred choice was difference in end-point data; however, when a (estimated unadjusted) MD between intervention and control at follow-up could not be calculated, the difference in change scores from baseline to follow-up was presented instead.

We had planned to undertake a mixed-treatment comparison of the multiple treatment options;[72] however, this was not possible owing to the paucity of RCTs for interventions other than melatonin. Adverse event data are summarised narratively.

Narrative synthesis

Although we planned to undertake separate quantitative and qualitative syntheses, this was to a large extent not possible, as few studies could be pooled. With the exception of the melatonin trials, there was a large degree of variability between studies evaluating different classes of interventions. For example, for

behavioural interventions, there was wide variability in aspects of the interventions such as mode of delivery, duration and intensity of interventions, as well as in the comparators used. There was also variability in the conditions being studied, outcomes reported and the measures used to assess individual outcomes, follow-up times and types of data reported. Consequently, there were few instances in which it became appropriate to pool the data. Thus, it became necessary to adopt a principally narrative synthesis to report the findings. We present a narrative synthesis as our main analysis, with comparisons made for melatonin versus placebo, which were the only interventions that we judged could be appropriately compared.

Narrative synthesis was undertaken when quantitative synthesis was not appropriate or there were insufficient data, and applied mainly to the non-pharmacological interventions. When possible, we display outcomes in a forest plot, even when studies are not statistically pooled, to aid exploration of study results. When feasible, we investigated the subgroup characteristics outlined previously. Non-pharmacological studies were grouped by type of intervention (i.e. comprehensive parent-directed tailored, comprehensive parent-directed non-tailored or non-comprehensive) and comparator if heterogeneous. We explored outcomes by type of sleep disturbance with the aim of identifying effects that may be transferable to other NDs. Results are discussed in the context of ratings of risk of bias in the individual studies.

In terms of the qualitative data analysis, the topic areas that were subject to review were well defined; we therefore adopted a thematic approach to data extraction, analysis and synthesis.[60,73] To start, studies were grouped into pharmacological, behavioural and other non-pharmacological studies. For each, a descriptive report of relevant studies, and topic areas covered, was produced. The tabulated data were then scrutinised and analytical notes were made that summarised findings across studies with respect to the topic areas. Part of this process involved testing for contradictions in the evidence.[74]

The synthesis interrogated such data, when available, to assist in identifying interventions that could be generalisable across conditions and those that are condition specific.[75,76] Factors taken into consideration in identifying promising interventions included feasibility of delivery of the intervention in a NHS setting, acceptability to children and families, evidence of clinical effectiveness or in the direction of clinical effectiveness based on CIs (taking into consideration the clinical significance of the estimates).

Protocol changes

During the course of the review, we identified numerous RCTs for the melatonin studies. We therefore decided to include only RCTs for this intervention. For all other interventions, we have included any design except case studies, as per the original protocol. Some studies reported multiple case studies that were not eligible for inclusion in the review. We made the decision to exclude uncontrolled studies with fewer than 10 participants.

Patient and public involvement

Three parents of children with NDs (two mothers, one father) acted as project advisors. They were recruited from a permanent parent consultation group of the chief investigator's research unit. These parents were invited to the project team meetings, which were held three times over the course of study and were attended by the research team and all co-applicants. Each parent attended at least one meeting. They were also consulted, via e-mail, regarding the implications of the findings of the review. The children's diagnoses included autism and rare, genetic conditions. At the first meeting, an early item on the agenda was a presentation of an overview of systematic reviews as a research method. Throughout the meetings, the parents were encouraged to share their experiences and opinions, and their contributions provided useful contextual information.

Chapter 3 Results

Study selection

The searches identified 23,292 records: 21,529 records were identified from the original searches undertaken in February and March 2016, 1563 were identified from the updated searches undertaken in February 2017, 194 were identified from trial registries and 6 were identified from subsequent reference checking (*Figure 1*). After removing duplicate references, 15,745 titles were screened. On the basis of titles, 14,420 titles were excluded; a further 937 were excluded on reading the abstract.

Full-text articles were sought for the remaining 388 records. We could not obtain full-text articles for 30 of the records. Of these, 11 were conference abstracts that were not available in full text[22,77–86] and five were trial registry entries that were recorded as 'complete' but had no study results available.[87–91] We contacted the authors to check whether or not there were any publications from the trials and received one response that the authors did not intend to publish the results as fewer than 10 participants had been recruited

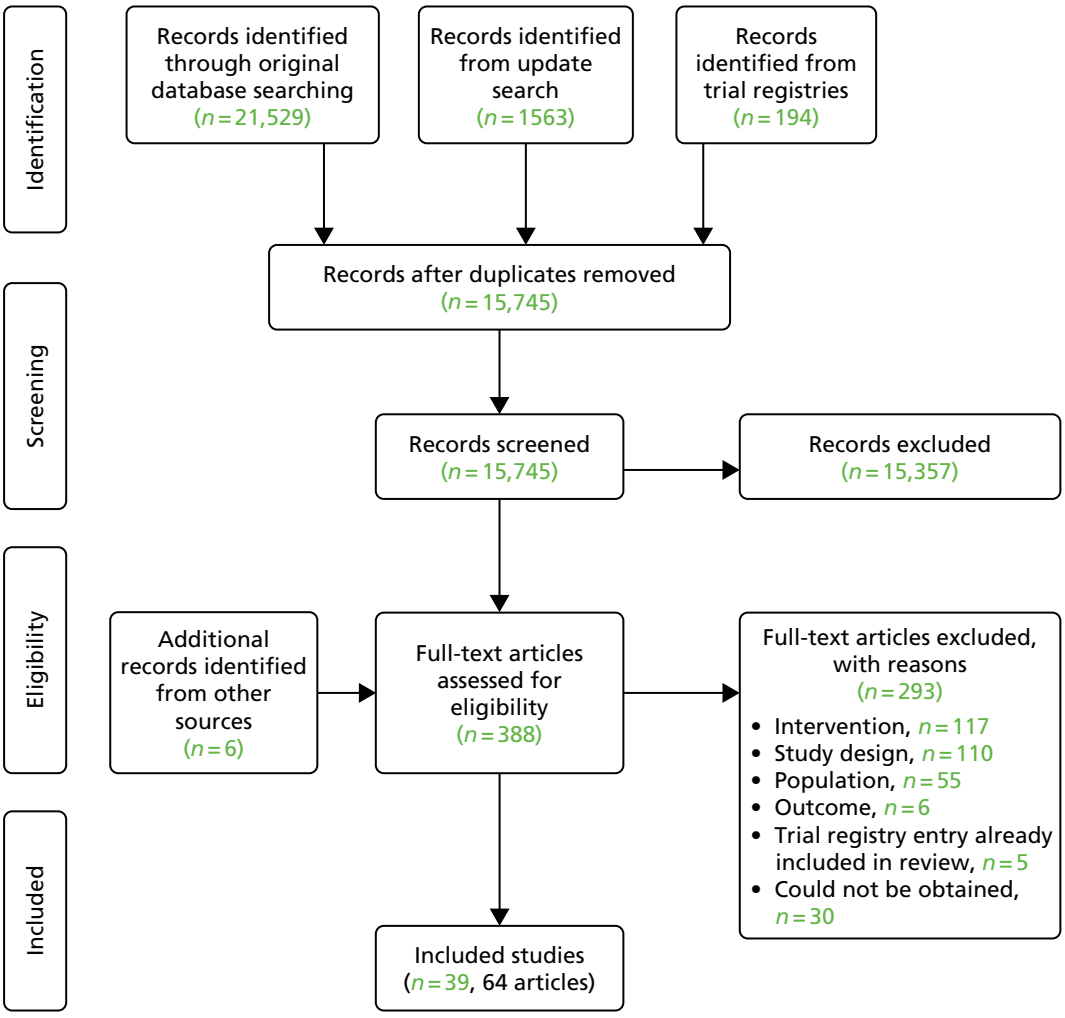

FIGURE 1 Flow chart of the study selection process.

into the study.[91] Two trials were registered as 'terminated' with no published results (one owing to poor recruitment,[92] and the other for unknown reasons [93] and our attempt to contact authors did not elicit a response). One trial was registered as 'study status unknown'[94] with no published results; we contacted the authors but received no response. Five trials were eligible but ongoing so were excluded from the review as results were not yet available.[95–99] A further six articles could not be obtained from the British Library.[100–105]

A total of 358 full-text articles were, therefore, assessed, including two non-English language papers that required translation. Of these, 117 articles (33%) were excluded because the intervention was out of scope or sleep disturbance was not the target of the intervention, 110 (31%) were excluded based on study design, 55 (15%) were excluded owing to the population, 6 (2%) were excluded based on the outcomes evaluated and 5 trials (1%) identified from trial registries were related to studies that had been completed and had already been identified through the searches and included in the review.[36,48,49,106,107] *Appendix 4* lists all studies excluded at full-text screening and reasons for exclusions.

Overview of included studies

There were 39 included studies reported in 64 articles. Although we sought to include studies published in any language, all the studies meeting the eligibility criteria were published in English. The included studies were from the UK ($n = 13$, 33%), the USA ($n = 10$, 26%), Australia ($n = 6$, 15%), Canada ($n = 5$, 13%), the People's Republic of China ($n = 1$, 2.6%), the Netherlands ($n = 1$, 2.6%), Hong Kong ($n = 1$, 2.6%), Italy ($n = 1$, 2.6%) and Israel ($n = 1$, 2.6%).

Thirteen RCTs investigated a pharmacological intervention, which in all cases was oral melatonin (*Table 1*). We did not identify any eligible pharmacological studies investigating clonidine or antihistamines. Twenty-six studies investigated non-pharmacological interventions, of which 12 were RCTs. Of these, nine (35%) evaluated parent-directed tailored interventions; eight (31%) evaluated parent-directed non-tailored interventions; two (8%) evaluated non-comprehensive parent-directed interventions; and seven (27%) evaluated other non-pharmacological interventions – dietary interventions ($n = 2$, 5%), alternative medicine ($n = 1$, 3%), exercise-based intervention ($n = 1$, 3%), faded bedtime with response costs ($n = 1$, 3%), weighted blankets ($n = 1$, 3%) and a light therapy plus a behavioural programme ($n = 1$, 3%). Thirteen studies explored feasibility and/or acceptability and/or parent/clinician views of sleep disturbance interventions.[36,49,106,107,122–130]

The mean age of the children included in the studies ranged from 2 to 12 years. In 16 studies (41%), participants were described in terms of a single ND diagnosis as follows: ADHD ($n = 5$), autism spectrum condition (ASC) ($n = 3$), ASD ($n = 3$), epilepsy ($n = 1$), tuberous sclerosis ($n = 1$), 'mental retardations' (we have replaced this term with 'learning disability'; this is interchangeable with the term 'intellectual disability') ($n = 2$) and severe ND ($n = 1$). In 22 studies (56%), participants were reported to have two or more NDs. One study did not report participants' NDs. There was also a range of sleep disturbance represented in the eligible studies. Owing to the different terminology used to describe sleep disturbances in the eligible studies, we have classified sleep disturbance under the following headings: sleep initiation ($n = 30$, 77%) (e.g. sleep latency, sleep association, settling, bedtime resistance and insomnia); sleep maintenance ($n = 26$, 67%) (e.g. night waking, waking time, parasomnia, co-sleeping and sleep fragmentations) and sleep scheduling ($n = 1$, 2.5%) (e.g. daytime sleepiness). Three studies (7%) reported that parents completed sleep questionnaires or assessment tools to determine their child's eligibility, including the Sleep Behaviour Questionnaire,[131] Quine sleep index[132] and the Children's Sleep Habits Questionnaire (CSHQ).[133] Five studies (13%) did not specify the types of sleep disturbance eligible for inclusion. Children often had multiple sleep disturbances.

TABLE 1 Characteristics of studies of melatonin interventions

Study details and design	Participants randomised (total *N* and by group)	Trial treatments	Sleep disturbance	Mean age (SD)	ND disorder	Previous sleep hygiene/ behavioural interventions	Risk of bias (low, unclear, high)
Melatonin vs. placebo: parallel-group RCTs							
Appleton *et al.* (2012)[48]							
Associated publications: Gringras *et al.* (2012),[108] Appleton *et al.* (2011),[109] Appleton *et al.* (2012)[48] UK	*N* = 146 (melatonin, *n* = 70; placebo, *n* = 76)	Melatonin: 0.5 mg, 45 minutes before bedtime for 12 weeks Placebo: matching capsule, 0.5 mg for 12 weeks Melatonin and placebo dose could be raised to 2 mg, 6 mg to 12 mg in first 4 weeks then maintained	Problems with sleep initiation and sleep maintenance	Melatonin: 106.0 months (34.8 months) Placebo: 100.7 months (37.4 months)	DD alone, and DD + other	Yes	Low
Cortesi *et al.* (2012)[110]							
Italy	*N* = 160 (melatonin, *n* = 40; melatonin and CBT, *n* = 40; CBT, *n* = 40; placebo, *n* = 40)	Melatonin: controlled release, 3 mg at 21.00 hours for 12 weeks CBT: four, weekly individual sessions Melatonin and CBT: as above Placebo: identical tablet, 3 mg at 21.00 hours for 12 weeks	Problems with sleep initiation and sleep maintenance	Melatonin: 6.8 years (0.9 years) Melatonin and CBT: 6.4 years (1.1 years) CBT: 7.1 years (0.7 years) Placebo: 6.3 years (1.2 years)	ASD	None stated	High
Van der Heijden *et al.* (2007)[111]							
Associated publications: Hoebert *et al.* (2009)[112] The Netherlands	*N* = 107 (melatonin, *n* = 54; placebo, *n* = 53)	Melatonin: fast release, 3 mg (if < 40 kg), 6 mg (if > 40 kg) at 19.00 hours for 4 weeks Placebo: identical appearing placebo at 19.00 hours for 4 weeks	Problems with sleep initiation	Melatonin: 9.1 years (2.3 years) Placebo: 9.3 years (1.8 years)	ADHD	None stated	Unclear

continued

TABLE 1 Characteristics of studies of melatonin interventions (*continued*)

Study details and design	Participants randomised (total *N* and by group)	Trial treatments	Sleep disturbance	Mean age (SD)	ND disorder	Previous sleep hygiene/ behavioural interventions	Risk of bias (low, unclear, high)
Crossover trials							
Camfield *et al.* (1996)[113]							
Individualised '*N* of 1 crossover trials' Canada	*N* = 6	Melatonin: 0.5 mg for three cases, 1.0 mg for three cases, taken at 18.00 hours Placebo: identical capsule Ten-week trial. For each of the five 2-week intervals, participants were randomised to receive placebo or melatonin for 1 week with the alternate agent given in the second week	Problems with sleep initiation and sleep maintenance	7.3 years (4.6 years)	Mixed	Yes	High
Dodge and Wilson (2001)[114]							
Associated publications: Hoebert *et al.* (2009)[112] USA	*N* = 36	Melatonin: 5 mg at 20.00 hours for 2 weeks Placebo: capsule and filler packaged to be identical to the melatonin, 5 mg at 20.00 hours for 2 weeks Washout period: 1 week	'Chronic sleep problems'	89 months (NR)	Mixed (mainly cerebral palsy)	Yes	Unclear
Garstang and Wallis (2006)[69]							
UK	*N* = 11	Melatonin: 5 mg for 4 weeks Placebo: dose NR, for 4 weeks Washout period: 1 week	Problems with sleep initiation and sleep maintenance	8.6 years (3.1 years)	ASD only, and ASD + learning disability	Yes	High

Study details and design	Participants randomised (total *N* and by group)	Trial treatments	Sleep disturbance	Mean age (SD)	ND disorder	Previous sleep hygiene/ behavioural interventions	Risk of bias (low, unclear, high)
Jain *et al.*[115] (2015)							
Associated Publications: Jain *et al.* (2014)[116]	*N* = 11	Melatonin: sustained release, 9 mg 30 minutes before bedtime for 4 weeks	A score of > 30 on the Sleep Behaviour Questionnaire	8.4 years (1.3 years)	Epilepsy	None stated	High
USA		Placebo: identical appearance to melatonin tablets, dose NR					
		Washout period: 1 week					
Wasdell *et al.* (2008)[117]							
Associated publications: Carr *et al.* (2007)[118]	*N* = 51	Melatonin: controlled release, 5 mg, 20–30 minutes before bedtime for 10 days	Problems with sleep initiation and sleep maintenance	7.4 years (NR)	Mixed	Yes	Unclear
Canada		Placebo: identical to melatonin, 5 mg, 20–30 minutes before bedtime for 10 days					
		'Placebo washout': 3–5 days					
Weiss *et al.* (2006)[70]							
Canada	*N* = 23	Melatonin: short-acting, 5 mg, 20 minutes before bedtime for 10 days	Problems with sleep initiation	10.3 years (NR)	ADHD	Yes	Unclear
		Placebo: for 10 days					
		'Placebo washout': 5 days					

continued

TABLE 1 Characteristics of studies of melatonin interventions (*continued*)

Study details and design	Participants randomised (total *N* and by group)	Trial treatments	Sleep disturbance	Mean age (SD)	ND disorder	Previous sleep hygiene/ behavioural interventions	Risk of bias (low, unclear, high)
Wirojanan *et al.* (2009)[119]							
USA	*N* = 18	Melatonin: 3 mg, 30 minutes before bedtime for 2 weeks Placebo: 3 mg, 30 minutes before bedtime for 2 weeks No washout period	'Sleep problem'	5.5 years (3.6 years)	Mixed (fragile X Syndrome/ASD)	None stated	Unclear
Wright *et al.* (2011)[106]							
UK	*N* = 20	Melatonin: standard release, 2 mg, 30–40 minutes before bedtime for 3 months Placebo: identical to melatonin, 2 mg, 30–40 minutes before bedtime for 3 months Melatonin and placebo dose increased by 2 mg every 3 nights to a maximum of 10 mg. Taken for 3 months Washout period: 1 month	Problems with sleep initiation and sleep maintenance	9.0 years (2.9 years)	Mixed (autism and Asperger syndrome)	Yes	Unclear

Study details and design	Participants randomised (total *N* and by group)	Trial treatments	Sleep disturbance	Mean age (SD)	ND disorder	Previous sleep hygiene/ behavioural interventions	Risk of bias (low, unclear, high)
Melatonin vs. melatonin-crossover trials							
Hancock *et al.* (2005)[120]							
UK	*N* = 8 (≤ 18 years, *n* = 5)	Melatonin: 1 × 5 mg plus 1 × 5-mg placebo, 30 minutes before bedtime for 2 weeks Melatonin: 2 × 5 mg of melatonin (10 mg in total), 30 minutes before bedtime for 2 weeks Washout period: 2 weeks	Quine sleep index score of at least 6	All: 12.1 years (10.0 years) ≤ 18 years: 6.9 years (4.0 years)	Tuberous sclerosis	None stated	High
Jan *et al.* (2000)[121]							
Canada	*N* = 16 (≤ 18 years, *n* = 15)	Melatonin: sustained release, variable doses from 2 mg to 10 mg, 30 minutes before bedtime for 11 days Control: fast-release melatonin, variable doses from 2 mg to 10 mg for 11 days No washout period	'Chronic sleep wake disorders'	All: 10.1 years (4.9 years) ≤ 18 years: 9.3 years (4.1 years)	Mixed	None stated	High

CBT, cognitive–behavioural therapy; DD, developmental delay; NR, not reported.

Melatonin

Study characteristics

Table 1 is a summary of the characteristics of the melatonin studies, and *Appendix 5* provides further details. Of the 13 RCTs evaluating oral melatonin, which were undertaken in Canada, Italy, the Netherlands, the UK and the USA, 10 compared melatonin with placebo only; one compared melatonin, cognitive–behavioural therapy and a combination of the two with placebo; and two compared two regimens of melatonin (5 mg vs. 10 mg, and fast release vs. sustained release). Ten were crossover trials and three were parallel-group RCTs. Observed sample sizes ranged from 6 to 160 participants.

Assessment of risk of bias

A summary assessment of risk of bias for each RCT is provided in *Table 1* and the full risk-of-bias assessment involving each bias domain is provided in *Appendix 6* and *Report Supplementary Material 1*. One trial was rated as having a low risk of bias.[48] The ratings of risk of bias in the remaining RCTs were high or unclear; therefore, the findings from these studies may not be robust.

We could not locate a registered protocol for nine trials[69,70,106,110,113–115,117,119] and one trial protocol was registered retrospectively,[111] making it unclear whether or not the studies were free of selective reporting. Seven studies provided no, or little, detail regarding sequence generation;[70,106,111,113,114,117,119] and four studies provided little or no detail regarding allocation concealment.[110,113,114,119] For three studies, how blinding was undertaken was unclear.[69,70,113] In three studies, the analysis of incomplete outcome data was not considered[69,110] or was unclear.[114]

Melatonin versus placebo

Eleven trials ($n = 589$ randomised participants) compared melatonin with placebo: eight crossover trials,[69,70,106,113–115,117,119] two two-armed parallel-group trials,[48,111] and one four-armed trial of oral melatonin, cognitive–behavioural therapy, oral melatonin plus cognitive–behavioural therapy and placebo.[110]

The washout period used by the crossover trials varied. One had no washout period,[119] one had a 1-month washout period[106] and the remaining five trials had a washout period of between 3 and 7 days.[69,70,114,115,117] Camfield *et al.*[113] reported six '*N* of 1' crossover trials with no washout period.

Six of the trials varied drug dosages depending on the child's age and/or weight and tolerance of dosage (see *Table 1*). The dose ranges were 0.5–1 mg,[113] 0.5–12 mg,[48] 3–6 mg[111] and 2–10 mg.[106] Fixed dosages were 3 mg ($n = 2$),[110,119] 5 mg ($n = 4$)[69,70,114,117] and 9 mg.[115] Matched placebos were used except for in one trial,[70] in which the placebo was not explicitly described, although in this trial the authors reported that each patient received a blister card of 30 days' supply of medication.

The length of time that melatonin was prescribed for varied between studies: 10 days,[70,117] 2 weeks,[114,119] 4 weeks,[69,111,115] 5 weeks[113] and 12 weeks.[48,106,110]

The age of participants included in these trials ranged from 1 to 18 years, although the mean age across studies was broadly similar, ranging from 5.5 to 10.3 years (see *Table 1*). The ND that was represented varied. Four trials included children with a mixed range of neurodevelopmental disabilities,[48,113,114,117] three trials included only children with ASC,[69,106,110] two trials included children with ADHD,[70,111] one trial included children with ASD and/or fragile X syndrome[119] and one trial included children with epilepsy.[115]

The type of sleep disturbances reported in the participants also varied. Most trials had more than one criterion for inclusion in the trial and included children with a mix of sleep disturbances (see *Table 1*) relating to sleep initiation[48,69,70,106,110,111,113,117] and sleep maintenance.[48,69,106,110,113,117] However, two trials focused on a single sleep problem ('sleep onset insomnia'[111] and 'initial insomnia'[70]) and one trial required a score of > 30 in the Sleep Behaviour Questionnaire.[115] Two studies did not specify the type of sleep problems eligible for inclusion.[114,119]

Seven trials specified in their inclusion criteria that a preceding psychoeducational or behavioural sleep management intervention had to have been ineffective.[48,69,70,106,113,114,117] In five of these trials, guidance on sleep was provided as part of the trial in the form of a behaviour therapy advice booklet,[48] tailored sleep hygiene advice,[117] a sleep hygiene intervention,[70] a sleep hygiene advice leaflet[69] and behaviour management and parenting support.[106] In four of these trials, only children who continued to experience sleep problems were randomised.[48,70,106,117]

Across these 11 trials, 19 sleep-related outcomes were measured (see *Appendix 7*). The most commonly measured outcomes were total sleep time (TST) ($n = 11$), sleep onset latency (SOL) ($n = 10$), number of night wakings ($n = 6$) and sleep efficiency ($n = 5$). Additional outcomes included arousals, bedtime, CSHQ, difficulty falling asleep, sleep onset, time from drug to sleep, longest sleep episode, the Sleep Behaviour Questionnaire, percentage of sleep stages, wake after sleep onset (WASO), nights without awakening, wake-up time, naptime, moving time, 'interdaily stability', 'interdaily variability' and 'L5' (average activity during the least active 5 hours). Definitions of outcome measures can be found in *Appendix 7*.

All trials had follow-up periods that commenced immediately following the completion of the intervention: 10 days,[70,117] 2 weeks,[114,119] 4 weeks,[69,111,115] 10 weeks[113] or 12 weeks.[48,106,110]

Global measures and composite scores

Total sleep time

All 11 trials[48,69,70,106,110,111,113–115,117,119] ($n = 589$ randomised participants) assessed TST (see *Appendix 7*); four measured this using parent-reported sleep diaries only,[69,106,113,114] whereas the remaining seven[48,70,110,111,115,117,119] used both actigraphy and sleep diaries. However, of these, three report only actigraphy-measured TST data,[110,111,119] as the sleep diaries were used purely to inform, or verify, actigraphy data. This follows existing guidance on the interpretation of actigraphy data.[134] The remaining four trials[48,70,115,117] reported TST derived from both parent-completed sleep diaries and actigraphy data.

Data from seven trials using sleep diary-reported TST were pooled: six crossover trials with a washout period ($n = 122$ analysed participants),[69,70,106,114,115,117] and one parallel-group trial ($n = 110$).[48] Note that in the forest plot (*Figure 2*), the sample sizes presented count participants in crossover trials twice (as being in the melatonin and placebo groups), so the figures reported in the text and shown in the figures may not match for this reason. There was a statistically significant increase in sleep diary-reported TST with melatonin compared with placebo (pooled MD 29.6 minutes, 95% CI 6.9 to 52.4 minutes; $p = 0.01$; see *Figures 2* and *3*). Statistical heterogeneity was high ($I^2 = 97\%$) and this treatment effect is unlikely to be generalisable, although the effect estimates were all in the direction of benefit with melatonin. Heterogeneity was reduced when studies were stratified based on whether or not the study population was exclusively ASD or not (test for subgroup differences: $p < 0.001$; $I^2 = 99\%$); there was a pooled MD of 64.7 minutes (95% CI 58.8 to 70.7 minutes, $I^2 = 0\%$) for the studies of ASD ($n = 24$), and a smaller pooled MD of 15.9 minutes (95% CI 9.2 to 22.6 minutes, $I^2 = 31\%$) for the studies of mixed or other populations ($n = 208$). There was only a single study ($n = 9$) in which participants had no prior sleep hygiene or behavioural intervention limiting the usefulness of this subgroup analysis; the overall results did not substantially change with removal of this study (pooled MD 33.0 minutes, 95% CI 8.6 to 57.4 minutes, $n = 223$; *Figure 3*). When the single trial rated as having a low risk of bias is considered alone, the increase in sleep time with melatonin was 13.2 minutes (95% CI –13.3 to 39.7 minutes).[48]

The study of six '*N* of 1' trials in six participants also reported sleep diary TST but, owing to the unusual trial design, could not be included in the meta-analysis.[113] The authors reported no 'notable difference in sleep pattern or daytime behaviour between melatonin and placebo weeks'[113] and provided raw data for each participant. From this, we calculated the MD between melatonin and placebo for parent-reported TST to be 13.9 minutes in favour of the melatonin group (95% CI –6.8 to 34.6 minutes; $p = 0.14$).

Study first author	MD	SE	Melatonin total	Placebo total	Weight	MD IV, random, 95% CI
ASD						
Garstang (2006)[69]	65.4	3.1	7	7	15.5%	65.40 (59.32 to 71.48)
Wright (2011)[106]	52.3	13.4	17	17	13.0%	52.30 (26.04 to 78.56)
Subtotal (95% CI)			24	24	28.5%	64.73 (58.81 to 70.65)
Heterogeneity: $\tau^2 = 0.00$; $\chi^2 = 0.91$, df = 1 ($p = 0.34$); $I^2 = 0\%$						
Test for overall effect: $z = 21.43$ ($p < 0.00001$)						
Not ASD						
Appleton (2012)[48]	13.2	13.5	51	59	12.7%	13.20 (–13.26 to 39.66)
Dodge (2001)[114]	18	13.2	20	20	13.0%	18.00 (–7.87 to 43.87)
Jain (2015)[115]	11.3	3	9	9	15.5%	11.30 (5.43 to 17.18)
Wasdell (2008)[117]	31.2	7.8	50	50	14.7%	31.20 (15.91 to 46.49)
Weiss (2006)[70]	15	4.8	19	19	15.3%	15.00 (5.59 to 24.41)
Subtotal (95% CI)			149	157	71.5%	15.87 (9.15 to 22.59)
Heterogeneity: $\tau^2 = 17.61$; $\chi^2 = 5.80$, df = 4 ($p = 0.21$); $I^2 = 31\%$						
Test for overall effect: $z = 4.63$ ($p < 0.00001$)						
Total (95% CI)			173	181	100.0%	29.63 (6.91 to 52.35)
Heterogeneity: $\tau^2 = 855.84$; $\chi^2 = 181.18$, df = 6 ($p < 0.00001$); $I^2 = 97\%$						
Test for overall effect: $z = 2.56$ ($p = 0.01$)						
Test for subgroup differences: $\chi^2 = 114.43$, df = 1 ($p < 0.00001$); $I^2 = 99.1\%$						

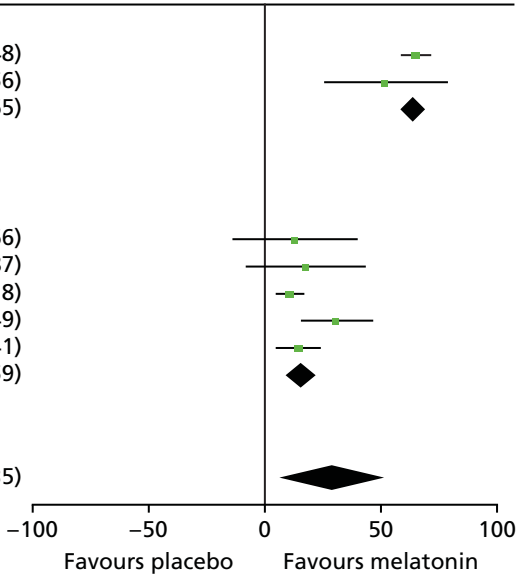

FIGURE 2 Sleep diary-reported TST: melatonin vs. placebo and ASD subgroup analysis. df, degrees of freedom; IV, instrumental variable.

Study first author	MD	SE	Melatonin total	Placebo total	Weight	MD IV, random, 95% CI
Prior intervention						
Appleton (2012)[48]	13.2	13.5	51	59	12.9%	13.20 (–13.26 to 39.66)
Dodge (2001)[114]	18	13.2	20	20	13.0%	18.00 (–7.87 to 43.87)
Garstang (2006)[69]	65.4	3.1	7	7	15.5%	65.40 (59.32 to 71.48)
Wasdell (2008)[117]	31.2	7.8	50	50	14.7%	31.20 (15.91 to 46.59)
Weiss (2006)[70]	15	4.8	19	19	15.3%	15.00 (5.59 to 24.41)
Wright (2011)[106]	52.3	13.4	17	17	13.0%	52.30 (26.04 to 78.56)
Subtotal (95% CI)			164	172	84.5%	33.01 (8.62 to 57.41)
Heterogeneity: $\tau^2 = 830.93$; $\chi^2 = 95.01$, df = 5 ($p < 0.00001$); $I^2 = 95\%$						
Test for overall effect: $z = 2.65$ ($p = 0.008$)						
No prior intervention						
Jain (2015)[115]	11.3	3	9	9	15.5%	11.30 (5.42 to 17.18)
Subtotal (95% CI)			9	9	15.5%	11.30 (5.42 to 17.18)
Heterogeneity: not applicable						
Test for overall effect: $z = 3.77$ ($p = 0.0002$)						
Total (95% CI)			173	181	100.0%	29.63 (6.91 to 52.35)
Heterogeneity: $\tau^2 = 855.84$; $\chi^2 = 181.18$, df = 6 ($p < 0.00001$); $I^2 = 97\%$						
Test for overall effect: $z = 2.56$ ($p = 0.01$)						
Test for subgroup differences: $\chi^2 = 2.88$, df = 1 ($p = 0.09$); $I^2 = 65.2\%$						

FIGURE 3 Sleep diary-reported TST: melatonin vs. placebo and prior intervention subgroup analysis. df, degrees of freedom; IV, instrumental variable.

Five trials[48,110,111,115,117] ($n = 266$ analysed participants) were pooled for actigraphy-measured TST, comprising two crossover trials with a washout period ($n = 60$)[115,117] and three parallel-group trials ($n = 206$).[48,110,111] Weiss *et al.*[70] used actigraphy to measure TST post intervention and reported that there was no significant difference between melatonin and placebo, but the study did not provide data on this outcome to allow it to be included in the meta-analysis.

There was a statistically significant increase in actigraphy-measured TST with melatonin compared with placebo (pooled MD 31.9 minutes, 95% CI 14.8 to 49.1 minutes; $p < 0.001$) (*Figure 4*). Heterogeneity was high ($I^2 = 76\%$) and this treatment effect is unlikely to be generalisable, although the effect estimates were all in the direction of benefit with melatonin. There was no statistically significant difference in effect between the studies in which participants received or did not receive a prior intervention (test for subgroup differences, $p = 0.48$; $I^2 = 0\%$). Subgroup analysis by type of ND was not possible.

One study without a washout period[119] ($n = 12$ analysed participants) reported a MD in actigraphy-measured TST post intervention of 21 minutes between melatonin and placebo, favouring melatonin. Using non-parametric analysis methods, the authors report *p*-values of 0.0019 and 0.02 for this outcome, based on data sets produced using two different approaches for dealing with missing data (complete case and last observation carried forward). This outcome was also analysed using a paired *t*-test, giving a *p*-value of 0.057. We have estimated the 95% CI for the MD of 21 minutes as –0.7 to 42.7 minutes.

For TST based on polysomnography at 4 weeks immediately post intervention,[115] there was no significant difference between melatonin and placebo, with a reported MD of 39.3 minutes (favouring placebo, 95% CI –34.7 to 113.3 minutes, $n = 10$).

Sleep efficiency

Five trials[48,110,111,115,117] ($n = 475$ randomised participants) reported sleep efficiency (i.e. the ratio of TST to total time in bed): one trial used both actigraphy and parent report,[117] three used actigraphy data only[48,110,111] and one trial used polysomnography.[115]

Cortesi *et al.*[110] also measured the percentage of children who achieved a sleep efficiency in the normative level of > 85% at the 12-week assessment.

Four trials[48,110,117,135] ($n = 255$ analysed participants) for actigraphy-measured sleep efficiency were pooled, comprising three parallel trials ($n = 205$) and one crossover trial with a washout period ($n = 50$). There was no statistically significant difference in sleep efficiency with melatonin compared with placebo (pooled MD 4.76% favouring melatonin, 95% CI –0.95% to 10.47%; $p = 0.10$; *Figure 5*). Heterogeneity was high ($I^2 = 94\%$) and this treatment effect is unlikely to be generalisable; although the trials did consistently report very small differences between groups in the proportion of time spent in bed asleep, these are unlikely to be clinically meaningful.

For the single trial[117] reporting parent-reported sleep efficiency ($n = 50$ analysed participants), there was no statistically significant difference between groups (MD 0.30% favouring melatonin, 95% CI –0.90% to 1.49%; $p = 0.62$).

Cortesi *et al.*[110] reported the percentage of children who achieved a sleep efficiency in the normative range (> 85%) as 46.43% of the melatonin group compared with none of the placebo group.

Jain *et al.*[115] reported no difference in polysomnography-measured sleep efficiency (MD 3.8% favouring melatonin, 95% CI –2.5% to 10.1%, $n = 10$).

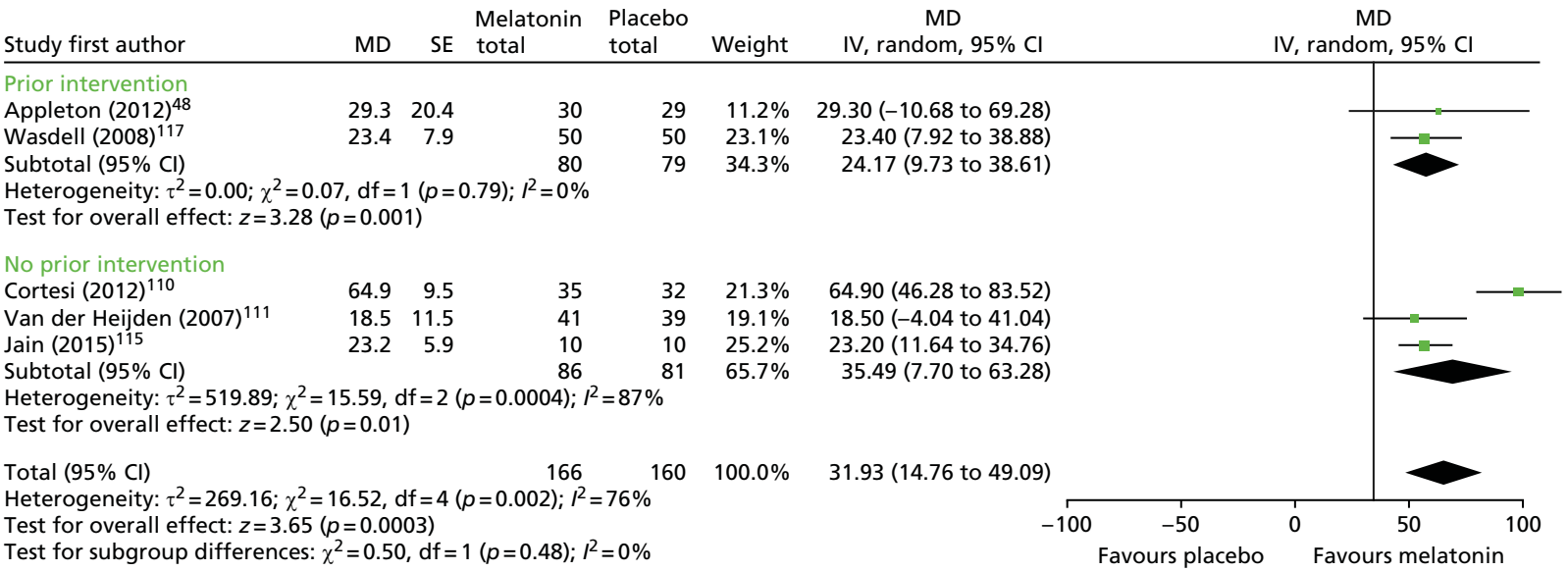

FIGURE 4 Actigraphy-measured TST: melatonin vs. placebo. df, degrees of freedom; IV, instrumental variable.

Study first author	MD	SE	Melatonin total	Placebo total	Weight	MD IV, random, 95% CI
Prior intervention						
Appleton (2012)[48]	5.4	3.02	30	28	21.2%	5.40 (–0.52 to 11.32)
Wasdell (2008)[117]	0.5	1.03	50	50	26.6%	0.50 (–1.52 to 2.52)
Subtotal (95% CI)			80	78	47.8%	2.13 (–2.40 to 6.65)
Heterogeneity: $\tau^2 = 6.91$; $\chi^2 = 2.36$, df = 1 ($p = 0.12$); $I^2 = 58\%$						
Test for overall effect: $z = 0.92$ ($p = 0.36$)						
No prior intervention						
Cortesi (2012)[110]	10.8	1.06	35	32	26.5%	10.80 (8.72 to 12.88)
Van der Heijden (2007)[111]	2.4	1.51	41	39	25.6%	2.40 (–0.56 to 5.36)
Subtotal (95% CI)			76	71	52.2%	6.67 (–1.56 to 14.90)
Heterogeneity: $\tau^2 = 33.58$; $\chi^2 = 20.73$, df = 1 ($p < 0.00001$); $I^2 = 95\%$						
Test for overall effect: $z = 1.59$ ($p = 0.11$)						
Total (95% CI)			156	149	100.0%	4.76 (–0.95 to 10.47)
Heterogeneity: $\tau^2 = 30.83$; $\chi^2 = 52.00$, df = 3 ($p < 0.00001$); $I^2 = 94\%$						
Test for overall effect: $z = 1.63$ ($p = 0.10$)						
Test for subgroup differences: $\chi^2 = 0.90$, df = 1 ($p = 0.34$); $I^2 = 0\%$						

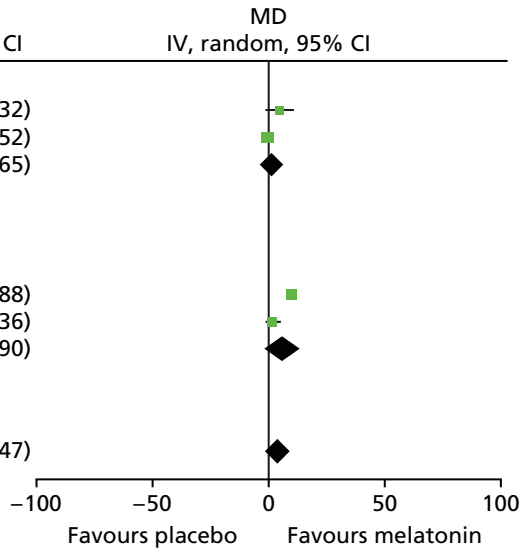

FIGURE 5 Actigraphy-measured sleep efficiency: melatonin vs. placebo. df, degrees of freedom; IV, instrumental variable.

Sleep initiation

Sleep onset latency

Ten trials[48,69,70,106,110,111,114,117,119] ($n = 583$ randomised participants) measured SOL, the time from bedtime to sleep onset time (see *Appendix 7*): three trials reported parent-reported SOL data only;[69,106,114] two reported actigraphy-measured SOL data only;[110,119] four reported both actigraphy-measured and parent-reported SOL data;[48,70,111,117] and one used polysomnography- and actigraphy-measured data, but reported only the results of the polysomnography.[115] As with TST, trials reporting actigraphy data stated that it was informed, or verified, by parent-reported sleep diaries.

Cortesi *et al.*[110] also calculated the percentage of children who met either a standard sleep criterion for SOL of ≤ 30 minutes or a reduction of SOL by 50%. Wright *et al.*[106] used an additional measure of SOL defined as the duration of time between taking the medication and falling asleep.

For parent-reported/sleep diary SOL, six trials[48,69,70,106,114,117] ($n = 223$ analysed participants) were pooled, comprising five crossover trials with a washout period ($n = 110$)[69,70,106,114,117] and one parallel-group trial ($n = 113$).[48] There was a statistically significant decrease (favouring melatonin) in SOL for melatonin compared with placebo (pooled MD –35.6 minutes, 95% CI –50.9 to –20.3 minutes; $p < 0.001$; *Figure 6*). Heterogeneity was high ($I^2 = 89\%$) and the treatment effect is unlikely to be generalisable, although the effect estimates were all in the direction of benefit with melatonin. There was a statistically significant difference in effect between the studies of children with ASD and those with mixed and other populations (test for subgroup differences, $p < 0.001$; $I^2 = 93\%$). There was a larger difference in the ASD group between melatonin and placebo, with a mean reduction in favour of melatonin of 50.9 minutes (95% CI –55.5 to –46.2 minutes) compared with 27.41 minutes (95% CI –39.1 to –15.7 minutes) in the other group. Subgroup analysis by whether or not participants had received a prior intervention was not possible.

For actigraphy-measured SOL, five trials[48,70,110,111,117] ($n = 265$ analysed participants) were pooled, comprising three parallel trials ($n = 196$)[48,110,111] and two crossover trials with a washout period ($n = 69$).[70,117] There was a statistically significant decrease (favouring melatonin) in actigraphy-reported SOL (pooled MD –23.4 points, 95% CI –30.9 to –15.8 points; $p < 0.001$; *Figure 7*). There was moderate heterogeneity ($I^2 = 48\%$). Based on the subgroup analysis, there was no statistically significant difference in effect between studies in which participants had or did not have a prior intervention (test for subgroup differences, $p = 0.55$; $I^2 = 0\%$). A subgroup analysis based on neurodevelopmental condition was not possible.

For both sleep diary-measured and actigraphy-measured SOL, the single study rated as having a low risk of bias[48] reported a statistically significant improvement with melatonin compared with placebo (see *Figures 6* and *7*).

The crossover study without a washout period[119] ($n = 12$ analysed participants) reported a statistically significant decrease in mean SOL for the melatonin period, compared with the placebo period, using a non-parametric analysis (complete-case analysis, $p = 0.02$; last observation carried forward, $p = 0.0001$), but this difference is not significant when tested using a paired *t*-test (MD –28.08 minutes; $p = 0.10$; estimated 95% CI –2.5 to 58.7 minutes).

Cortesi *et al.*[110] used an additional indicator of SOL (the percentage of children who met a criterion of SOL of ≤ 30 minutes or a reduction of SOL by 50% post intervention) and reported that 39% of the melatonin group versus 0% of the placebo group achieved these changes ($n = 66$ analysed participants). Jain *et al.*[115] ($n = 10$), using polysomnography, reported that melatonin significantly reduced the mean SOL compared with placebo, with a reported MD of –11.4 minutes (95% CI –17.2 to –5.6 minutes).

Wright *et al.*[106] ($n = 17$) reported a significant decrease in SOL with melatonin compared with placebo [MD 51.7 minutes (SD 71.9 minutes; $p = 0.012$; estimated 95% CI 16.5 to 86.9 minutes)].

Study first author	MD	SE	Melatonin total	Placebo total	Weight	MD IV, random, 95% CI
ASD						
Garstang (2006)[69]	−51	2.4	7	7	20.8%	−51.00 (−55.70 to −46.30)
Wright (2011)[106]	−46.7	13.3	17	16	13.1%	−46.70 (−72.77 to −20.63)
Subtotal (95% CI)			24	23	34.0%	−50.86 (−55.49 to −46.24)
Heterogeneity: $\tau^2 = 0.00$; $\chi^2 = 0.10$, df = 1 ($p = 0.75$); $I^2 = 0\%$						
Test for overall effect: $z = 21.54$ ($p < 0.00001$)						
Not ASD						
Appleton (2012)[48]	−37.5	10.9	54	59	15.0%	−37.50 (−58.86 to −16.14)
Dodge (2001)[114]	−30	14	17	17	12.6%	−30.00 (−57.44 to −2.56)
Wasdell (2008)[117]	−33.4	5.9	50	50	18.9%	−33.40 (−44.96 to −21.84)
Weiss (2006)[70]	−15.7	5.1	19	19	19.5%	−15.70 (−25.70 to −5.70)
Subtotal (95% CI)			140	145	66.0%	−27.41 (−39.13 to −15.69)
Heterogeneity: $\tau^2 = 74.44$; $\chi^2 = 6.79$, df = 3 ($p = 0.08$); $I^2 = 56\%$						
Test for overall effect: $z = 4.59$ ($p < 0.00001$)						
Total (95% CI)			164	168	100.0%	−35.55 (−50.85 to −20.26)
Heterogeneity: $\tau^2 = 286.69$; $\chi^2 = 43.60$, df = 5 ($p < 0.00001$); $I^2 = 89\%$						
Test for overall effect: $z = 4.56$ ($p < 0.00001$)						
Test for subgroup differences: $\chi^2 = 13.31$, df = 1 ($p = 0.0003$); $I^2 = 92.5\%$						

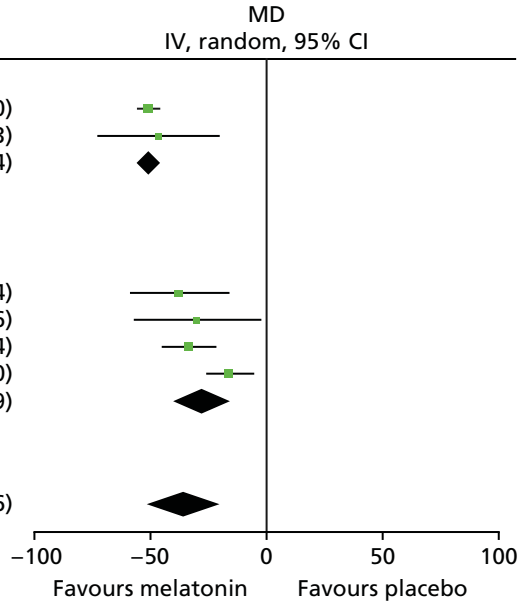

FIGURE 6 Sleep diary-measured SOL: melatonin vs. placebo. df, degrees of freedom; IV, instrumental variable.

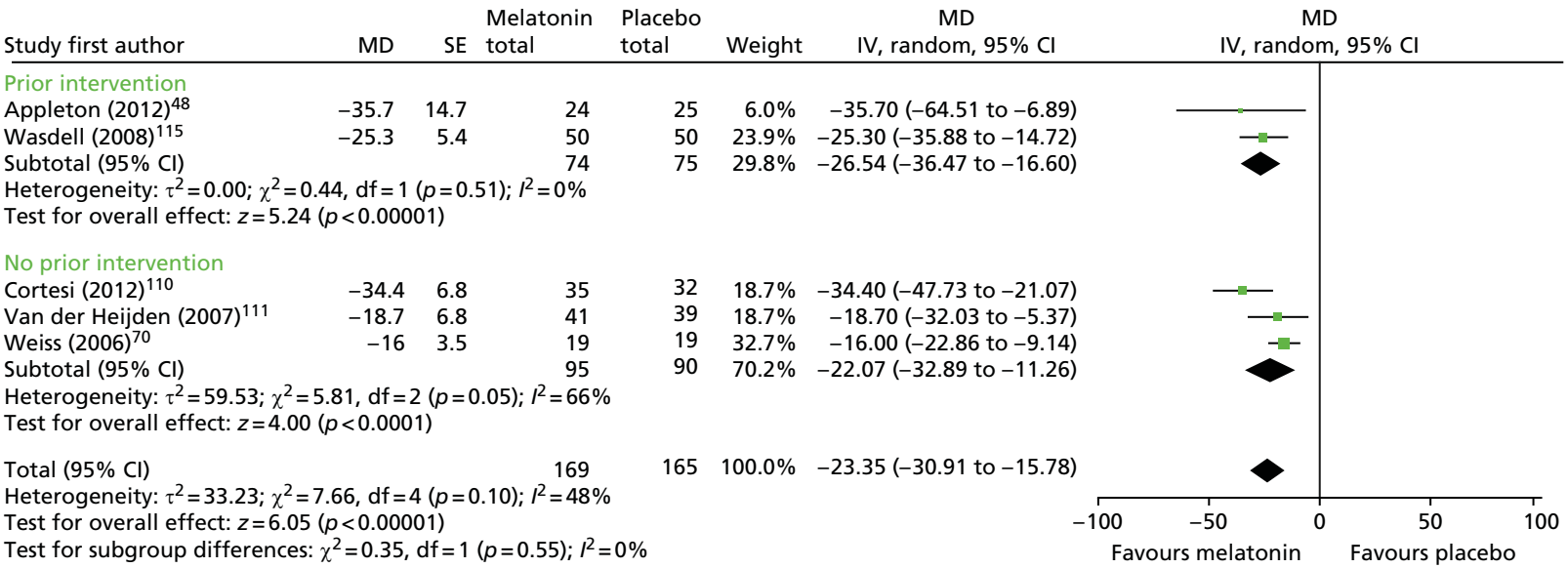

FIGURE 7 Actigraphy-measured SOL: melatonin vs. placebo. df, degrees of freedom; IV, instrumental variable.

Sleep maintenance

Number of night wakings

Six trials[69,106,113,114,117,119] ($n = 142$ randomised participants) reported the number of night wakings.

Four crossover trials with a washout period were pooled ($n = 94$ analysed participants).[69,106,114,117] There was no difference in the mean number of night wakings with melatonin compared with placebo (pooled MD –0.04 points, 95% CI –0.22 to 0.13 points; $p = 0.61$; *Figure 8*). Heterogeneity was high ($I^2 = 84\%$), although the results were fairly consistent, with the exception of those of Dodge and Wilson.[114] Based on the subgroup analysis, there was no statistically significant difference in effect between studies based on ND (test for subgroup differences, $p = 0.06$; $I^2 = 71\%$).

The crossover study without a washout period[119] reported no significant difference for the melatonin period compared with the placebo period by either the non-parametric analyses or the paired *t*-test [MD by paired *t*-test –0.07 points (favours melatonin, $p = 0.73$; estimated 95% CI –0.44 to 0.30 points)]. Wasdell *et al.*[117] reported no statistically significant treatment difference ($p = 0.48$) for melatonin compared with placebo (estimated MD –0.41 points favouring melatonin, 95% CI –1.47 to 0.66 points).

Other outcomes

Six out of the 11 trials reported other outcomes of interest,[110,111,113,115,117,119] although, with the exception of WASO (night waking duration and/or frequency after the child falls asleep), which was reported by two studies,[110,115] each outcome measure was reported by only a single study. These data are summarised in *Table 2*.

Melatonin versus melatonin

Two crossover trials[120,121] ($n = 24$ randomised participants) compared melatonin with melatonin. Jan *et al.*[121] compared controlled-release with fast-release melatonin, and Hancock *et al.*[120] compared a dose regimen of 5 mg with a dose regimen 10 mg of melatonin.

Jan *et al.*[121] did not have a washout period between the two intervention phases, whereas Hancock *et al.*[120] had a 2-week washout period. Hancock *et al.*[120] compared a fixed-dose regimen of 5 mg with 10 mg of melatonin regardless of the child's age. The authors did not report whether or not the melatonin was controlled or fast release. The final average dose for controlled-release melatonin for Jan *et al.*[121] was 5.7 mg (range 2–12 mg), and the final average dose for fast-release melatonin was 7 mg (range 2–15 mg). Jan *et al.*[121] reported that the dosage for fast-release melatonin varied for each child depending on the dosage deemed by the trial team to be most effective for that child. However, the controlled-release melatonin was approximately 50% of the fast-release dose, which the authors stated was to avoid possible adverse side effects, as they had no previous information on the use of controlled-release melatonin in children. The duration of the intervention was 11 days for Jan *et al.*[121] and 2 weeks for Hancock *et al.*[120]

The age of participants included in the trials ranged from 18 months to 31 years for Hancock *et al.*[120] (eligible participants < 18 years, $n = 5$, mean age 6.9 years); and from 4 to 21 years for Jan *et al.*[121] (eligible participants < 18 years, $n = 15$, mean age 9.3 years). Children in Hancock *et al.*[120] had severe neurodevelopmental difficulties, whereas the ND in the Jan *et al.*[121] study was tuberous sclerosis. The sleep disturbance in both studies focused on both sleep initiation and maintenance: in Hancock *et al.*[120] 'severe sleep problems', confirmed by a Quine sleep index score of at least 6 out of a possible 8, and for Jan *et al.*[121] 'chronic wake–sleep disorders'.

In Jan *et al.*,[121] the inclusion criterion was that children had already been treated with fast-release melatonin for > 3 months but slept for < 5–6 hours. No additional guidance or leaflets on sleep management were provided to parents in either of the trials.

Study or subgroup	MD	SE	Melatonin total	Placebo total	Weight	MD IV, random, 95% CI
ASD						
Garstang (2006)[69]	−0.18	0.02	7	7	30.8%	−0.18 (−0.22 to −0.14)
Wright (2011)[106]	−0.1	0.1	17	17	22.2%	−0.10 (−0.30 to 0.10)
Subtotal (95% CI)			24	24	53.0%	−0.18 (−0.22 to −0.14)
Heterogeneity: $\tau^2 = 0.00$; $\chi^2 = 0.62$, df = 1 ($p = 0.43$); $I^2 = 0\%$						
Test for overall effect: $z = 9.02$ ($p < 0.00001$)						
Not ASD						
Dodge (2001)[114]	0.2	0.09	20	20	23.5%	0.20 (0.02 to 0.38)
Wasdell (2008)[115]	−0.06	0.09	50	50	23.5%	−0.06 (−0.24 to 0.12)
Subtotal (95% CI)			70	70	47.0%	0.07 (−0.18 to 0.32)
Heterogeneity: $\tau^2 = 0.03$; $\chi^2 = 4.17$, df = 1 ($p = 0.04$); $I^2 = 76\%$						
Test for overall effect: $z = 0.54$ ($p = 0.59$)						
Total (95% CI)			94	94	100.0%	−0.04 (−0.22 to 0.13)
Heterogeneity: $\tau^2 = 0.02$; $\chi^2 = 18.54$, df = 3 ($p = 0.0003$); $I^2 = 84\%$						
Test for overall effect: $z = 0.51$ ($p = 0.61$)						
Test for subgroup differences: $\chi^2 = 3.53$, df = 1 ($p = 0.06$); $I^2 = 84\%$						

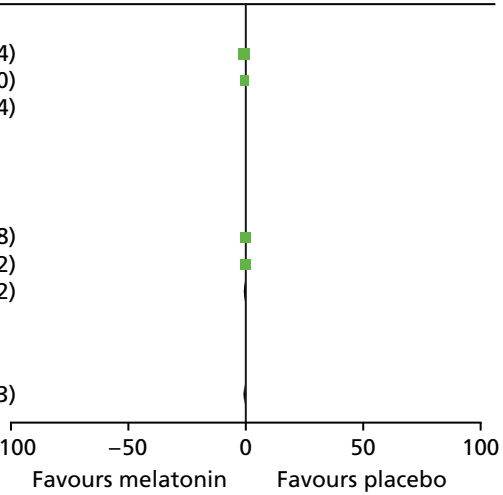

FIGURE 8 Parent-reported number of night wakings: melatonin vs. placebo. df, degrees of freedom; IV, instrumental variable.

TABLE 2 Other outcome results from studies of pharmacological interventions

Study	Outcome	Time point, mean (SD)		MD between melatonin and placebo (95% CI)
		Baseline	Follow-up	
Global measures and composite scores				
Cortesi *et al.* (2012)[110]	CSHQ (total points)	Melatonin: 66.67 (8.55)	Melatonin only: 54.78 (6.22)	–10.02 (–12.71 to –7.33)
		Placebo: 64.20 (4.85)	Placebo: 64.80 (4.52)	
	WASO (minutes)	Melatonin: 73.71 (45.00)	Melatonin: 42.21 (22.35)	–27.94 (–44.58 to –11.30)
		Placebo: 69.75 (45.21)	Placebo: 70.15 (42.76)	
Jain *et al.* (2015)[115]	Sleep Behaviour Questionnaire (total points)	Overall: 59.2 (10.5)	Melatonin: 52.4 (5.4)	3.4 (–2.2 to 9.0)
			Placebo: 48.3 (7.4)	
	WASO (minutes)	Overall: 60.6 (24.0)	Melatonin: 42.3 (30.3)	–22.2 (–34.1 to –10.3)
			Placebo: 57.3 (31.6)	
Wasdell *et al.* (2008)[117]	Somnolog longest sleep episode (minutes)	Overall: 415.41 (106.23)	Melatonin: 453.30 (118.41)	18.3 (–5.0 to 41.6)
			Placebo: 434.26 (109.09)	
	Actigraph longest sleep episode (minutes)	Overall: 185.17 (102.63)	Melatonin: 199.37 (100.46)	7.9 (–16.6 to 32.4)
			Placebo: 189.25 (99.98)	
Sleep initiation				
Cortesi *et al.* (2011)[7]	Bedtime (units unclear)	Melatonin: 23.45 (1.15)	Melatonin: 22.30 (1.10)	–1.21 (–1.76 to –0.66)
		Placebo: 23.41 (1.19)	Placebo: 23.51 (1.12)	
Van der Heijden *et al.* (2007)[111]	Sleep onset (hour : minutes)	Melatonin: 21:40 (0.59 minutes)	Melatonin: 21:13 (0:58)	–35 minutes (–59 to –11 minutes)
			Placebo: 21:48 (0:48)	
		Placebo: 21:38 (0.47 minutes)		
	Difficulty falling asleep[a] (points)	Melatonin: 3.4 (0.9)	Melatonin: 2.2 (0.9)	–0.9 (–1.3 to –0.5)
		Placebo: 3.2 points (0.7 points)	Placebo: 3.1 points (1.0 points)	
Jain *et al.* (2015)[115]	Bedtime (hour : minutes)	Overall: 21:44 (0.75 minutes)	Melatonin: 21:57 (0.91 minutes)	6.8 minutes (–23.3 to 9.7 minutes)
			Placebo: 21:49 (0.72 minutes)	
Wirojanan *et al.* (2009)[119]	Sleep onset time (hour : minutes)	NR	Melatonin: 20:43 (1.39 minutes)	–42 minutes (–74.8 to –9.2 minutes)
			Placebo: 21:25 (2.00 minutes)	

TABLE 2 Other outcome results from studies of pharmacological interventions (*continued*)

Study	Outcome	Time point, mean (SD)		MD between melatonin and placebo (95% CI)
		Baseline	Follow-up	
Sleep maintenance				
Camfield *et al.* (1996)[113]	Nights without awakening	NR	Raw data for nights without awakening between 22:00 and 07:00 hours per day/the number of days of complete data: Melatonin: 2/25 (8%); 0/21 (0%); 7/35 (20%); 10/29 (34%); 4/33 (12%); 26/35 (74%) Placebo: 2/28 (7%); 0/25 (0%); 1/31 (3%); 4/30 (13%); 1/31 (3%); 24/35 (69%)	8.9 (–0.1 to 17.9)
Van der Heijden *et al.* (2007)[111]	Wake up time[b] (hour : minutes)	Melatonin: 07:25 (0.39) Placebo: 07:25 (0.34)	Melatonin: 07:21 (0.40 minutes) Placebo: 07:33 (0.26 minutes)	–12.0 minutes (–27.1 to 3.1 minutes)
	Moving time[b] (%)	Melatonin: 11.95 (4.38) Placebo: 10.43 (3.69)	Melatonin: 12.79 (8.20) Placebo: 12.30 (3.88)	0.5 (2.4 to 3.4)
Jain *et al.* (2015)[115]	Wake time (hour : minutes)	Overall: 7:09 (1:04)	Melatonin: 7:31 (1.09 minutes) Placebo: 7:11 (1.09 minutes)	–18.1 minutes (–0.2 to –36.0 minutes)
Sleep scheduling				
Cortesi *et al.* (2012)[110]	Naptime (units unclear)	Melatonin: 33.57 (56.63) Placebo: 37.33 (56.19)	Melatonin: 17.00 (33.11) Placebo: 36.10 (33.28)	–19.10 (–35.43 to –2.77)
Other outcomes				
Van der Heijden *et al.* (2007)[111]	Interdaily stability[b,c] (points)	Melatonin: 0.65 (0.13) Placebo: 0.64 (0.15)	Melatonin: 0.66 (0.16) Placebo: 0.68 (0.11)	–0.02 (–0.08 to 0.04)
	Intradaily variability[b,d] (points)	Melatonin: 0.65 (0.18) Placebo: 0.67 (0.15)	Melatonin: 0.69 (0.23) Placebo: 0.63 (0.14)	0.06 (–0.03 to 0.15)
	L5 (average activity during the least active 5 hours) (points)	Melatonin: 44.89 (26.72) Placebo: 36.01 (27.09)	Melatonin: 39.57 (28.86) Placebo: 50.56 (28.67)	–11.0 (–24.0 to 2.0)

continued

TABLE 2 Other outcome results from studies of pharmacological interventions (*continued*)

Study	Outcome	Time point, mean (SD)		MD between melatonin and placebo (95% CI)
		Baseline	Follow-up	
Jain *et al.* (2015)[115]	Arousal index per hour of sleep	Overall: 12.9 (7.4)	Melatonin: 10.3 (4.1)	2.5 (–2.5 to 7.5)
			Placebo: 12.3 (6.8)	
	Percentage of sleep stages (%)			
	N1	Overall: 6.3 (2.9)	Melatonin: 4.0 (2.0)	–0.1 (–0.5 to 0.3)
			Placebo: 4.8 (2.8)	
	N2	Overall: 42.6 (5.2)	Melatonin: 44.0 (9.2)	–2.9 (–10.5 to 4.7)
			Placebo: 42.6 (8.1)	
	N3	Overall: 28.9 (3.4)	Melatonin: 32.6 (8.3)	–5.5 (–7.6 to –3.4)
			Placebo: 27.4 (4.8)	
	REM stage (%)	Overall: 22.2 (3.7)	Melatonin: 19.4 (4.2)	5.8 (4.2 to 7.4)
			Placebo: 25.2 (5.1)	
	REM latency (minutes)	Overall: 136.2 (43.6)	Melatonin: 135.2 (53.1)	–58.3 (–36.2 to –80.4)
			Placebo: 98.1 (52.8)	

NR, not reported; REM, rapid eye movement.
a Mean difficulty falling asleep as reported by parents, averaged over 7 days on a scale of 1 (not difficult) to 5 (very difficult).
b Data available from online supplementary material, not reported in paper.
c Range from 0 to 1, with higher values indicating more stable rhythms.
d Range from 0 to 2, with higher values indicating more fragmented rhythms.

Across both trials, four sleep-related outcomes were measured (see *Appendix 8*). Hancock *et al.*[120] measured TST, SOL and number of night wakings, whereas Jan *et al.*[121] measured 'changes in sleep pattern' as their only outcome. The results of these outcomes are summarised in *Table 3* for participants aged < 18 years. Each outcome measure was reported by a single study. There was no evidence of benefit on any of the sleep-related outcomes for the RCT that compared controlled-release melatonin with fast-release melatonin[121] or the RCT that compared a dose of 5 mg of melatonin with 10 mg of melatonin.[120]

Adverse events

Eleven out of the 13 trials measured adverse events,[48,70,106,110,111,114,115,117,119–121] which were collected and reported in different ways across the studies (*Table 4*).

One study reported adverse event data using the standardised assessment tool 'treatment-emergent signs and symptoms', which classified events into seven domains: somnolence, increased excitability, mood swings, seizures, rash, hypothermia and cough.[48] Signs and symptoms were graded as 'no symptoms', 'mild symptoms', 'moderate symptoms' and 'severe symptoms'. Seriousness and causality were also assessed. In two studies,[110,115] adverse events were determined at each in-person/telephone call visit by the study team and recorded in the participant's chart; one study measured adverse events using study-specific questionnaires completed by parents;[106] one reported using a standardised form;[70] one reported using parent-completed questionnaires, in which the origin of the questionnaire was not reported;[114] two reported measuring adverse events using open-ended interviews;[111,117] one study reported only that adverse effects were reported by parents;[120] and in one study it was unclear how the data were collected.[119]

TABLE 3 Outcome results from trials comparing melatonin with melatonin

Study	Outcome	Time point, mean (SD)		MD between interventions (95% CI)
		Baseline	Follow-up	
Global measures and composite scores				
Hancock *et al.* (2005)[120]	TST (minutes)	NR	Melatonin, 5 mg: 548.6 (18.9) Melatonin, 10 mg: 548.0 (34.6)	–0.6 (–35.8 to 34.6)
Jan *et al.* (2000)[121]	Changes in sleep pattern	N/A	Improvements in the sleep patterns, to the satisfaction of the caregivers, were observed in 10 participants during the controlled-release period relative to the fast-release period	N/A
Sleep initiation				
Hancock *et al.* (2005)[120]	SOL (minutes)	NR	Melatonin, 5 mg: 70.0 (58.0) Melatonin, 10 mg: 68.4 (46.9)	–1.6 (–37.4 to 34.2)
Sleep maintenance				
Hancock *et al.* (2005)[120]	Number of night wakings	NR	Melatonin, 5 mg: 0.8 (0.4) Melatonin, 10 mg: 1.0 (0.9)	0.3 (–0.7 to 1.2)

N/A, not applicable; NR, not reported.

TABLE 4 Adverse events in studies evaluating melatonin

Study	AEs	Method of collection
Appleton *et al.* (2012)[48]	Melatonin: mild AEs, $n = 151$; moderate AEs, $n = 35$; severe AEs, $n = 3$ (waking up in the night because of nightmares, severe irritation to skin, seizure) Other non-treatment-emergent signs and symptoms: fatigue, $n = 8$; headache, $n = 10$; other, $n = 31$. Seizures: pre randomisation, $n = 49$; post randomisation, $n = 211$ Placebo: mild AEs, $n = 195$; moderate AEs, $n = 28$; severe AEs, $n = 4$ (dislocated elbow in accident at school, petechiae covering the dorsum of the right hand, choking on dinner, vomiting caused by viral illness, which caused dehydration) Other non-treatment-emergent signs and symptoms: fatigue, $n = 8$; headache, $n = 7$; other, $n = 40$. Seizures: pre randomisation, $n = 61$; post randomisation, $n = 192$	Treatment-emergent signs and symptoms tool
Camfield *et al.* (1996)[113]	Not reported	Not reported
Cortesi *et al.* (2012)[110]	No AEs were reported or observed	Recorded during face-to-face visits/telephone calls by study team

continued

TABLE 4 Adverse events in studies evaluating melatonin (*continued*)

Study	AEs	Method of collection
Van der Heijden *et al.* (2007)[111] Associated paper: Hoebert *et al.* (2009)[112]	Melatonin: one AE, $n = 5$, two AEs, $n = 4$, three AEs, $n = 1$. There were no discontinuations or withdrawals as a result of AEs. None of the AEs required treatment. At 3 weeks post intervention: headache, $n = 3$; hyperactivity, $n = 3$; dizziness, $n = 2$; abdominal pain, $n = 2$; nose bleeding, $n = 1$; itching lumps on the skin, $n = 1$; painful lumps on the skin, $n = 1$; diarrhoea, $n = 1$; decrease of mood, $n = 1$; maintenance of insomnia, $n = 1$. At 2 years after participation: 7 out of 24 parents reported one or more AE: bedwetting, $n = 2$, abnormal faeces, $n = 2$; drowsiness, $n = 2$; dizziness, $n = 1$; sleep maintenance problems, $n = 1$; skin pigment changes, $n = 1$; decreased mood, $n = 1$ Placebo: no AEs reported Long-term follow-up (mean follow-up time 3.66 years) with 94 parents of children who participated in the Van der Heijen *et al.*[111] trial: 19 children experienced AEs that they or their parents attributed to melatonin treatment. The majority of parents (63.2%) reported multiple AEs; seven reported one AE; four reported two AEs; four reported three AEs and four parents reported four AEs. Ten children had AEs that were self-limiting. In six children the AEs persisted and were a reason to discontinue treatment in three out of six children. In three children it was not mentioned in the questionnaire if the AEs were self-limiting or not. Dizziness, $n = 4$; visual disturbances, $n = 2$; melatonin treatment bedwetting, $n = 3$; excessive morning sedation, $n = 2$; sleep maintenance insomnia, $n = 3$; constipation, $n = 1$; headache, $n = 2$; profuse perspiration, $n = 1$; nausea, $n = 2$; decreased mood, $n = 1$; skin pigment changes, $n = 2$; daytime laziness, $n = 1$; nightmares, $n = 2$; and change in behaviour, $n = 1$	Open-ended interviews Parent-completed questionnaire
Dodge and Wilson (2001)[114]	Melatonin: more moody, $n = 1$; more 'hyper', $n = 1$. No reported changes in seizure frequency Placebo: more moody, $n = 1$; more 'hyper', $n = 1$ (different child). No reported changes in seizure frequency	Parent-completed questionnaire
Garstang and Wallis (2006)[69]	Not reported	Not reported
Hancock *et al.* (2005)[120]	No adverse effects were reported during the trial Seizures ($n = 5$). There was no change in the frequency (or type) of seizures seen compared with the baseline period before melatonin treatment at either dose	'Reported by parents'
Jain *et al.* (2015)[115]	Melatonin: AEs ($n = 4$); increased severity of headache ($n = 1$). 'Unrelated adverse events': bronchitis and ear infection ($n = 1$), agitation ($n = 1$) and increased urinary frequency ($n = 1$ continued from placebo phase) Placebo: AEs $n = 2$. 'Unrelated adverse events': agitation ($n = 1$ continued from melatonin phase) and increased urinary frequency ($n = 1$)	Recorded during face-to-face visits/telephone calls by study team
Jan *et al.* (2000)[121]	No adverse effects were experienced during the trial	Not explicitly stated – response to melatonin was measured through sleep charts and parental history

TABLE 4 Adverse events in studies evaluating melatonin (*continued*)

Study	AEs	Method of collection
Wasdell *et al.* (2008)[117] Associated paper: Carr *et al.* (2007)[118]	98 AEs reported across arms Melatonin: 36%. Most common AEs: seizures, $n = 11$; cold/flu/infection, $n = 8$; gastrointestinal illness, $n = 5$; agitation, $n = 4$; anxiety, $n = 2$ – considered consistent with patient's medical history and not related to melatonin; headache, $n = 2$ – considered consistent with patient's medical history and not related to melatonin. 40% of AEs in treatment group were from one patient: seizures, agitation, gagging and headaches, which were considered consistent with patient's medical history 'Treatment with controlled release melatonin was well tolerated and no treatment differences were evident on vital signs or physical examinations'[117] Placebo: 40% (and 24% in placebo washout phases) Most common AEs: cold/flu/infection, $n = 10$; seizures, $n = 8$; gastrointestinal illness, $n = 5$; and behavioural problems (agitation, anxiety, irritability, emotional lability), $n = 7$. One serious AE consistent with participant's medical history (aspiration pneumonia requiring hospitalisation) occurred during placebo treatment in the first period of the crossover trial After 3-month open-label phase: 16 AEs reported, consistent with pre-existing medical conditions. One serious AE: patient admitted to hospital for 3 days owing to an upper respiratory infection Prospective long-term, open-label follow-up of melatonin: mean (SD), frequency of AEs: nausea, 0.15 (0.65), 0–4; vomiting, 0.05 (0.22), 0–1; diarrhoea, 0.10 (0.37), 0–1; impaired appetite, 0.05 (0.31), 0; weight loss, 0.05 (0.31), 0–2; confusion, 0 (0), 0; excessive morning sedation, 0.12 (0.51), 0–3; depression, 0.10 (0.63), 0–4; irritability, 0.27 (0.71), 0–3; hyperactive behaviour, 0.10 (0.37), 0–2; deterioration of behaviour, 0.10 (0.49), 0–3; regression of development, 0 (0), 0; precocious puberty, 0 (0), 0; increase in seizures, 0.07 (0.26), 0–1; nasal allergy, 0.12 (0.40), 0–2; rash, 0.12 (0.46), 0–2; deterioration in asthma, 0.07 (0.35), 0–2; worsening of balance, 0.02 (0.16), 0–1; new tremor, 0.05 (0.22), 0–1; headache, 0.07 (0.35), 0–2; visual disturbance, 0.02 (0.16), 0–1; interference with other medications, 0 (0), 0; interference with other therapies, 0 (0), 0	Open-ended interviews Telephone call with caregivers
Weiss *et al.* (2006)[70]	Melatonin: 20% of all AEs reported during the melatonin phase Placebo: 23% of all AEs reported during the placebo phase All mild/moderate adverse effects with exception of one severe AE (migraine). Rash from actigraph ($n = 2$) No serious AEs and no clinically significant changes in vital signs or abnormalities on physical examination	Recorded on a standardised form that included severity, timing and relationship to the study drug
Wirojanan *et al.* (2009)[119]	No side effects were reported by parents	Unclear

continued

TABLE 4 Adverse events in studies evaluating melatonin (*continued*)

Study	AEs	Method of collection
Wright *et al.* (2011)[106]	Authors report that there were no statistically significant differences between treatment and placebo in the frequencies of reported side effects (but do not give actual numbers), including the following: daytime drowsiness, dizziness, headaches, vomiting, tummy aches, reduced appetite, low mood, anxiety, irritability, reduced alertness, confusion, tearfulness, diarrhoea, constipation, rashes, sort throat, ear aches, asthma, fit/seizure, mild tremor and 'other'. Daytime drowsiness, reduced appetite, reduced alertness and diarrhoea were reported as 'never present' more often in the placebo than treatment group, but the difference was not statistically significant. There were no serious AEs One child stopped medication during an influenza episode and did not continue as sleep 'continued to be good'. There were no known reports of seizures or asthma during the trial. One child displayed increased moodiness and self-injurious behaviour but clinicians reported that these were long-standing problems for the child	Study-specific questionnaire

AE, adverse event.

Three trials reported that no adverse events were observed or reported during the trial.[110,119,121] Further details of the adverse events reported can be found in *Table 4*. Overall, melatonin was tolerated and the adverse event profile appeared to be similar between the melatonin and placebo groups.

Summary

Eleven RCTs (eight crossover and three parallel trials) compared melatonin, of varying doses and duration, with placebo. The mean age of participants ranged from 5.5 to 10.3 years and some studies included children with a single ND, whereas others included mixed populations. Children had mostly sleep initiation and/or sleep maintenance problems and in the majority of studies had been offered a prior intervention, ranging from a sleep hygiene advice leaflet to behaviour management and parenting support. Only three child-related sleep outcomes were reported by more than half of the studies, with many of the additional outcomes assessed by a single study. None of the studies assessed parent-related outcomes.

One study was assessed as having a low risk of bias, with the remaining studies having an unclear or high risk of bias. When possible, studies were grouped in the synthesis by ND (ASD vs. mixed or other groups) and by whether or not they had received a prior intervention. However, the results of the subgroup analyses should be interpreted with caution, rather than be considered definitive, as they are based on summary data and are, therefore, observational rather than randomised comparisons as in a trial. In addition, some of the subgroups contained a particularly small number of studies.

There was evidence of benefit with melatonin compared with placebo, although the precise extent of the benefit, which children might benefit the most and the clinical importance of the benefit remain uncertain. The overall benefit was similar for diary-reported and actigraphy-recorded TST. Based on two small studies of children with ASD, there was a mean increase in diary-recorded TST of 64.78 minutes with melatonin compared with placebo (95% CI 58.8 to 70.7 minutes); for the five mixed or other ND studies, the mean increase was 15.9 minutes (95% CI 9.2 to 22.6 minutes). For the single study that was rated as having a low risk of bias (mixed population), the mean increase in TST was 13.2 minutes and the lower CI included the possibility of reduced sleep time (95% CI –13.3 to 39.7 minutes). The findings were similar for SOL. The benefit was greatest for the ASD studies in which there was a mean reduction of 50.9 minutes (95% CI –55.5 to –46.2 minutes) in diary-measured SOL, based on two small studies, whereas there was a smaller mean reduction of 27.4 minutes (95% CI –39.1 to –15.7 minutes) for the mixed and other population subgroup. For the single study with a low risk of bias (mixed population), the mean reduction in SOL was 37.5 minutes (95% CI –58.9 to –16.1 minutes). For other outcomes, there was some evidence

of statistically significant benefit with melatonin for sleep initiation outcomes but not sleep maintenance outcomes, such as number of night wakings, although some of the single studies may not have been sufficiently powered to detect an effect.

A single RCT[121] compared controlled-release and fast-release melatonin and one RCT[120] compared a 5 mg and a 10 mg dose of melatonin, both with a high risk of bias. There was no evidence of benefit with either strategy.

Non-pharmacological studies

Study characteristics

Twenty-six studies evaluated non-pharmacological interventions: 12 RCTs, one controlled before-and-after study and 13 uncontrolled before-and-after studies. Sample sizes ranged from 5 to 244 participants. Studies were conducted in Australia, Canada, China, Hong Kong, Israel, the UK and the USA. A summary of key characteristics is provided in *Table 5* and further details are provided in *Appendix 9*.

We grouped studies, based on the intervention, into the following: parent-directed tailored interventions ($n = 9$), parent-directed non-tailored interventions ($n = 8$), non-comprehensive parent-directed interventions ($n = 2$) and 'other' non-pharmacological interventions ($n = 7$). Intervention and control details for non-pharmacological studies are outlined in *Appendix 10*.

Parent-directed interventions can be conceived as psychoeducational interventions that have the objective of 'training' parents to manage their child's sleep disturbance by equipping them with the relevant knowledge and skills. These interventions vary in their intensity and different modes are used to deliver the training, including individual work, group work, teaching workshops and written material. Nineteen studies[21,49,107,123–129,138,146,149,151,152,155,162,163] of parent-directed interventions were included in the review. The interventions varied considerably in terms of a number of characteristics. There was no consistency in the terminology used by study authors to describe the interventions, and the terms that were used to describe an intervention were not routinely defined. This was not unexpected. Generally, reporting of non-pharmacological interventions is acknowledged to be much poorer than for pharmacological studies.[164] However, it carries the risk of erroneous comparison of interventions or pooling of studies.

Therefore, a framework to describe the parent-directed interventions represented in this study was developed by the research team. It included intervention characteristics posited as being an 'active ingredient' of an intervention and/or having an impact on intervention effectiveness, for example, intensity, duration, mode of delivery and whether the intervention was condition specific or ND generic. There was incomplete reporting on these intervention characteristics across the studies. We defined tailored and non-tailored interventions as follows:

- Tailored – a face-to-face clinical assessment by a trained practitioner guides clinical 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 extended 'implementation support', that is, ongoing support and advice as the parents implement changes to sleep management strategies and practices. The intervention is typically delivered on a one-to-one basis. However, an intervention that used teaching workshops and one-to-one work was also categorised as tailored[123,124] because it fulfilled the criteria defined above.
- Non-tailored – the delivery of a standard 'training curriculum'. The curriculum may include opportunities for a parent to be supported to operationalise the material learnt for their child's sleep disturbance. Some implementation support may also be included. Among the studies included in this review, such interventions were delivered in a number of ways: written material, single-session workshops, group delivery and one-to-one work.

TABLE 5 Study details for studies evaluating non-pharmacological interventions

Study details and design	Participants randomised (total *N* and by group)	Intervention details	Sleep disturbance	Mean age (SD)	ND disorder	Previous sleep hygiene/ behavioural interventions	Risk of bias (low, unclear, high)
Parent-directed tailored interventions: RCTs							
Beresford *et al.* (2012)[21] UK Associated publications: Beresford *et al.*, (2013)[136] Stuttard *et al.* (2015)[45,137] Parallel RCT	*N* = 13 Intervention group: *n* = 7 Comparison group: *n* = 6	Intervention group: two face-to-face sessions for assessment, development of sleep management strategy and training parent in strategy. Implementation support delivered via telephone calls Comparison group: usual approach to providing sleep management intervention – as above but implementation support delivered via home visits	Problems with sleep initiation and maintenance	Intervention group: 2.86 years (0.82 years) Comparison group: 2.67 years (1.07 years)	Mixed	None stated	High
Hiscock *et al.* (2015)[138] Australia Associated publications: Papadopoulos *et al.* (2015)[139] Parallel RCT	*N* = 244 Intervention group: *n* = 122 Comparison group: *n* = 122	Intervention group: one session for assessment, development of sleep management strategy and training parent in strategy. Implementation support delivered via one face-to-face session and one telephone call Comparison group: usual care	Problems with sleep initiation	Intervention group: 10.3 years (1.8 years) Comparison group: 9.9 years (2.1 years)	ADHD + LD or ASD/Asperger syndrome	None stated	High[a]
Johnson *et al.* (2013)[107] USA Associated publications: Turner (2013)[140] Parallel RCT	*N* = 40 Intervention group: *n* = 20 Comparison group: *n* = 20	Intervention group: one session for assessment, development of sleep management strategy; five sessions training parent in strategy. Implementation support delivered via one face-to-face session Comparison group: non-sleep-related parent education delivered in identical manner to intervention group	Problems with sleep initiation and maintenance	Intervention group: 3.5 years (0.98 years) Comparison group: 3.6 years (1.12 years)	Autism and ASD	None stated	High

Study details and design	Participants randomised (total *N* and by group)	Intervention details	Sleep disturbance	Mean age (SD)	ND disorder	Previous sleep hygiene/ behavioural interventions	Risk of bias (low, unclear, high)
Moss *et al.* (2014)[124]							
Australia Associated publications: O'Connell *et al.* (2012 and 2010)[141,142] Parallel RCT	*N* = 26 Intervention group: *n* = 13 Comparison group: *n* = 13	Intervention group: two training workshops followed by a home visit for assessment and development of sleep-managed strategy. Implementation support delivered via one home visit followed by telephone calls as needed Comparison group: waiting list control	Problems with sleep initiation, maintenance and scheduling and snoring	11.74 years (2.53 years) (NR separately)	Mixed	One child taking melatonin	High
Sciberras *et al.* (2011)[125]							
Australia Associated publications: Sciberras *et al.*, (2010)[143] Sciberras and Rinehart, (2015)[144] Fulton *et al.* (2010)[145] Parallel RCT	*N* = 27 Intervention group: *n* = 14 Comparison group: *n* = 13	Intervention group: two sessions for assessment, development of sleep management strategy and training parent in the strategy. Implementation support delivered via a single telephone call followed by a further face-to-face session if needed Comparison group: single session for assessment, development of sleep management strategy and training parent in the strategy. No implementation support	Problems with sleep initiation	Intervention group: 12.1 years (2.2 years) Comparison group: 10.9 years (2.5 years)	ADHD	None stated	High
Parent-directed tailored interventions: before-and-after studies							
Austin *et al.* (2013)[123]							
Australia	*N* = 8	Two training workshops followed by a home visit for assessment and development of sleep management strategy, followed by a third workshop. Implementation support delivered via weekly telephone calls	Problems with sleep initiation and maintenance	4.0 years (1.9 years)	Mixed	None stated	High

continued

TABLE 5 Study details for studies evaluating non-pharmacological interventions (*continued*)

Study details and design	Participants randomised (total *N* and by group)	Intervention details	Sleep disturbance	Mean age (SD)	ND disorder	Previous sleep hygiene/ behavioural interventions	Risk of bias (low, unclear, high)
Beresford *et al.* (2013)[21]							
UK Associated publications: Beresford *et al.*, (2013)[136] Stuttard *et al.* (2015)[45,137] Intervention 2	*N* = 12	Two sessions for assessment, development of sleep management strategy and training parent in strategy. Implementation support delivered via fortnightly face-to-face sessions	Problems with sleep initiation and maintenance	2.88 years (1.25 years)	Mixed	None stated	High
Quine and Wade (1991)[146]							
UK Associated publications: Wade and Wade (1991)[147]	*N* = 25	Two sessions for assessment, development of sleep management strategy and training parent in strategy. Implementation support delivered via face-to-face sessions	Problems with sleep initiation and maintenance	Mean (SD) NR (range 3–21 years)	LD	None stated	High
Weiskop *et al.* (2005)[126]							
Australia Before and after with multiple baseline	*N* = 13	Four sessions for assessment, development of sleep management strategy and training parent in strategy. Implementation support (via telephone calls) delivered from outset of intervention, continuing after training sessions completed with a face-to-face session and further telephone calls	Problems with sleep initiation and maintenance	5.1 years (2.0 years)	Mixed	One child was taking medication for behaviour and sleep problems	High
Parent-directed non-tailored interventions: RCTs							
Adkins *et al.* (2012)[127]							
USA	*N* = 36	Intervention group: training curriculum contained in a booklet provided to parent	Problems with sleep initiation	6.4 years (2.6 years) (NR separately)	Mixed	None stated	High

Study details and design	Participants randomised (total *N* and by group)	Intervention details	Sleep disturbance	Mean age (SD)	ND disorder	Previous sleep hygiene/ behavioural interventions	Risk of bias (low, unclear, high)
Associated publications: Malow *et al.* (2011)[148] Parallel-group RCT	Intervention group: *n* = 18 Comparison group: *n* = 18	Comparison group: no booklet provided					
Montgomery *et al.* (2004)[49]							
UK Associated publications: Montgomery *et al.* (2004)[49] Parallel-group RCT	*N* = 66 Intervention group a: *n* = 22 Intervention group b: *n* = 34 Comparison group: *n* = 26	Intervention group a: training curriculum contained in a booklet provided to parent Intervention group b: training curriculum (identical to that contained in booklet) delivered via face-to-face session Comparison group: no intervention (waiting list)	Problems with sleep initiation and maintenance	Mean (SD) NR (range 27–101 months, NR separately)	Mixed	None stated	High
Malow *et al.* (2014)[128]							
USA Parallel-group RCT	*N* = 80 Intervention group: *n* = 39 Comparison group: *n* = 41	Intervention group: training curriculum delivered via two group-delivered sessions. Implementation support delivered via telephone calls Comparison group: training curriculum delivered via single face-to-face session. Implementation support delivered via telephone calls	Problems with sleep initiation	Intervention group: 5.9 years (2.8 years) Comparison group: 5.6 years (2.6 years)	Mixed	None stated	High
Parent-directed non-tailored interventions: before-and-after studies							
Beresford *et al.* (2012)[21]							
UK	*N* = 22	Group delivery of training curriculum over four sessions	Problems with sleep initiation	8.91 years (3.25 years)	Mixed	None stated	High

continued

TABLE 5 Study details for studies evaluating non-pharmacological interventions (*continued*)

Study details and design	Participants randomised (total *N* and by group)	Intervention details	Sleep disturbance	Mean age (SD)	ND disorder	Previous sleep hygiene/ behavioural interventions	Risk of bias (low, unclear, high)
Associated publications: Beresford *et al.* (2013)[136] Stuttard *et al.* (2015)[45,137]							
Intervention 3							
Beresford *et al.* (2012)[21]							
UK Associated publications: Beresford *et al.* (2013)[136] Stuttard *et al.* (2015)[45,137]	*N* = 25	Training curriculum delivered via a single half-day workshop	Problems with sleep initiation and maintenance	7.0 years (3.3 years)	Mixed	None stated	High
Intervention 4							
Bramble (1997)[149]							
UK Associated publications: Bramble (1996)[122]	*N* = 15	Training curriculum delivered via single session. Implementation support delivered via telephone calls	Problems with sleep initiation and maintenance	7.2 years (2.6 years)	Mixed	Previous interventions indicated but not described, other than as sedatives	High
Reed *et al.* (2009)[129]							
Canada Associated publications: Reed *et al.* (2008)[150]	*N* = 22	Group delivery of training curriculum over three sessions	Problems with sleep initiation and maintenance	5.8 years (2.7 years)	ASD	None stated	Unclear

Study details and design	Participants randomised (total *N* and by group)	Intervention details	Sleep disturbance	Mean age (SD)	ND disorder	Previous sleep hygiene/ behavioural interventions	Risk of bias (low, unclear, high)
Yu *et al.* (2015)[151]							
Hong Kong	*N* = 54	Group delivery of training curriculum over three sessions, supported by weekly telephone calls. Implementation support delivered via telephone calls	Problems with sleep initiation and maintenance	4.78 years (0.85 years)	ASD and Asperger syndrome	None stated	High
Non-comprehensive parent-directed interventions: RCTs							
Wiggs and Stores (1998)[152]							
UK Associated publications: Wiggs and Stores (1999 and 2001)[153,154] Cluster RCT	*N* = 30 Intervention group: *n* = 15 Comparison group: *n* = 15	Intervention group: tailored intervention. Single session for assessment, development of sleep management strategy and training parent in strategy. Implementation support delivered via telephone calls Comparison group: no intervention	Problems with sleep initiation and maintenance	Intervention group: 8.21 years (2.7 years) Comparison group: 10.77 years (3.8 years)	Mixed	None stated	High
Non-comprehensive parent-directed interventions: before-and-after study							
Peppers *et al.* (2016)[155]							
USA	*N* = 23	Intervention group: prescriptive sleep hygiene intervention. One session delivered by a practitioner	Global measure of sleep disturbance (CSHQ) used to define eligibility to receive intervention	Mean (SD) NR (range 5–11 years)	NR	None stated	High
Other interventions – RCTs							
Francis and Dempster (2002)[156]							
Australia Crossover RCT	*N* = 5	Intervention group: the intervention contained valerian of 500 mg per tablet, 30 mg per kg of body weight, single nightly dose ≥ 1 hour before bedtime, 2 weeks Comparison group: to give the same appearance and odour, placebo contained 25 mg of whole-root *Valeriana edulis* extract, 2 weeks	Problems with sleep initiation and maintenance	6.6 years (3.4 years)	Mixed	None stated	High

continued

TABLE 5 Study details for studies evaluating non-pharmacological interventions (*continued*)

Study details and design	Participants randomised (total *N* and by group)	Intervention details	Sleep disturbance	Mean age (SD)	ND disorder	Previous sleep hygiene/ behavioural interventions	Risk of bias (low, unclear, high)
Gringras *et al.* (2014)[36]							
UK Crossover RCT	*N* = 73	Intervention group: weighted blanket 2.25 kg (small) or 4.5 kg (large). 12–16 days and were given by researchers at home/clinic visits Comparison group: placebo blanket	Problems with sleep initiation and maintenance	Weighted blanket first: 8.7 years (3.3 years) Control blanket first: 9.9 years (2.8 years)	Mixed	None stated	High
Piazza *et al.* (1997)[157]							
USA Parallel-group RCT	*N* = 14 Intervention group: *n* = 7 Comparison group: *n* = 7	Intervention group: faded bedtime with response cost, 10 days. The study author delivered the face-to-face (home) visits and booklet intervention Comparison group: bedtime scheduling	Problems with sleep maintenance and initiation	Intervention group: 6.7 years (2.6 years) Comparison group: 8.3 years (3.0 years)	Mixed	Children were excluded if receiving pharmacological interventions for sleep. No other previous interventions reported	High
Other non-pharmacological interventions: non-randomised study designs							
Guilleminault *et al.* (1993)[158]							
USA	*N* = 14	Light therapy and behavioural programme. Daily light exposure at 07.00 and 12.00 hours	Problems with sleep maintenance and 'lack of sleep consolidation'	2.9 years (range 9 months to 4 years)	Moderate to severe learning disability	Eight children had attended sleep clinics and centres for treatment. Some children had had behavioural treatments	High

Study details and design	Participants randomised (total *N* and by group)	Intervention details	Sleep disturbance	Mean age (SD)	ND disorder	Previous sleep hygiene/ behavioural interventions	Risk of bias (low, unclear, high)
Oriel *et al.* (2016)[159]							
USA A–B–A withdrawal design	*N* = 8	Aquatic exercise programme. 60 minutes of aquatic exercise two times per week	Parent/guardian report of sleep dysfunction	8.9 years (SD NR, range 6–11 years)	ASD	None stated	High
Yehuda *et al.* (2011)[160]							
Israel Controlled before and after	*N* = 78 Intervention group: *n* = 40 Comparison group: *n* = 38 (Healthy control *n* = 22 not included)	Intervention group: essential fatty acids supplement, 90 g of α-linolenic and 360 g of linoleic acid in mineral oil. Two capsules per day for 10 weeks Comparison group: placebo	Sleep deprived	(All) mean (SD) NR (range 9–12 years)	ADHD	None stated	Unclear
Yu and Hong (2012)[161]							
China Before and after	*N* = 30	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 was given three times a week, with 36 sessions constituting one course. Two courses were required	Problems with sleep initiation, maintenance and abnormal sleep state (including apnoea)	6.9 years (3.1 years)	Learning disability	None stated	High

LD, learning disability; NR, not reported.

a This study was rated as having a low risk of bias for all domains except blinding, which was not possible owing to the nature of the intervention. However, in keeping with the guidance outlined in *Chapter 2*, *Assessment of risk of bias*, it has been rated as having a high risk of bias, as the subjective outcomes may have been influenced by the lack of blinding.

All but two of the parent-directed interventions were comprehensive in their content, that is, they included training across three topic areas: sleep and sleep processes, sleep hygiene and the management of specific problem behaviours (e.g. night wakings). The remaining two interventions, which were classified as non-comprehensive parent-directed interventions, focused on a single topic area related to managing sleep disturbance. One was concerned only with sleep hygiene training[155] and the other focused on particular behavioural strategies to manage specific problem behaviours.[152]

Assessment of risk of bias

A summary assessment of risk of bias for each RCT is provided in *Table 5* and the full risk-of-bias assessment involving each bias domain is provided in *Appendix 21*. Overall, the studies were rated as having a risk of bias that was high ($n = 24$) or unclear ($n = 2$); therefore, the findings from these studies may not be robust. Following the guidance outlined in *Chapter 2*, *Assessment of risk bias*, one study was rated as having a high risk of bias, as blinded outcome assessment was not possible.[138] However, this study had no other important limitations and all other domains were rated as having a low risk of bias.

We were unable to find a registered protocol for 10 RCTs,[21,36,49,107,124,125,127,128,152,156] making it unclear whether or not the studies were free of selective reporting. Eight trials provided little or no detail regarding sequence generation.[21,36,49,107,124,127,152,157] Ten RCTs provided little or no detail regarding allocation concealment.[21,36,49,107,124,125,127,128,152,156] In all RCTs, blinded outcome assessment was not undertaken[21,36,49,107,125,127,128,138,152] or it was unclear whether or not blinding occurred.[124,156] In eight RCTs, the analysis of incomplete outcome data was not considered[21,36,125,128,152] or it was unclear whether or not the authors had considered this.[107,124,127]

Eleven non-randomised designs[21,123,126,146,149,155,159–163] were reported as having a risk of bias owing to a probable or unclear risk of confounding of the effect of the intervention. Fourteen studies were reported as having a risk of bias because of how they selected participants, for example by not reporting inclusion and/or exclusion criteria, selecting participants from one setting only or including more males than females.[21,123,126,129,146,149,151,155,158–163] There was likely or unclear bias reported in the measurement of intervention outcomes in seven studies, for example few or no details provided about how outcomes were measured.[21,126,146,159–161] One further study[160] was reported as having unclear bias because of departures from intended interventions, owing to missing data and to the selection of the reported results.

Parent-directed tailored interventions

Nine studies evaluated parent-directed, comprehensive tailored interventions (*Table 6*). Seven studies evaluated the clinical effectiveness of these interventions: three two-armed parallel-group RCTs[107,124,138] with a range of no intervention comparators (i.e. usual care, waiting list control or non-sleep related parent education); and four before-and-after studies.[21,123,126,146] Two further RCTs[21,125] evaluated alternative ways of delivering an intervention. One compared the mode by which implementation support was provided: home visit versus telephone call.[21] The other compared the intensity of practitioner involvement when delivering the intervention: brief versus extended.[125]

Six studies included children with a range of neurodevelopmental conditions,[21,123,124,126,146] two included children with a diagnosis of ADHD[125,138] and one included children with a diagnosis of ASD.[107]

Interventions varied in terms of the total number of sessions delivered; the number of sessions spent in assessment; the development of, and training in, a sleep management plan; the duration of the intervention; and the mode of delivery (see *Table 6*). The number of assessment/training sessions ranged from one[21,138] to five[107] sessions in the RCTs; and from two[21,146] to four sessions[123,126] in the other study designs. In the RCTs, the intervention duration was one session,[125] 4 weeks[138] and 10 weeks,[21] and was not reported in two studies.[107,124] In the other studies,[21,123,126,146] the duration of the intervention ranged from 6 to 28 weeks.

TABLE 6 Details of parent-directed tailored interventions

Study	Total duration of intervention (including period of implementation support)	Mode of delivery	Number of sessions and location	Mode of delivering implementation support, and intensity, once regular sessions with practitioner were completed	Intervention described as developed for specific ND?	Manual?	Follow-up, from baseline
RCTs							
Hiscock *et al.* (2015)[138]	4 weeks	Face to face	One (home or clinic)	Face to face ($n = 1$), later followed by a telephone call ($n = 1$)	Yes, ADHD	No	3 and 6 months
Johnson *et al.* (2013)[107]	NR	Face to face	Five (home and clinic)	Face to face ($n = 1$)	Yes, ASD	Yes	1 and 2 months
Moss *et al.* (2014)[124]	15 weeks	Teaching workshops and face to face	Two workshops and one face-to-face session (home)	Home visit ($n = 1$), followed by telephone calls, 'on a needs basis for approximately 2 months'	No	Yes	15 and 23 weeks
Before-and-after studies							
Austin *et al.* (2013)[123]	15 weeks	Teaching workshops and face to face	Two workshops, one home visit and one workshop	Approximately weekly telephone call for 6-week period	No	Yes	19 weeks
Beresford *et al.* (2012)[21]	12–16 weeks	Face to face	Two (clinic, home)	Fortnightly sessions at clinic	No	No	12 and 24 weeks
Quine and Wade (1991)[146]	6–28 weeks	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	3 months
Weiskop *et al.* (2005)[126]	Minimum 7 weeks	Face to face	Four (a mixture of home and clinic), plus at least weekly telephone contact between sessions	'Review session' 5 weeks after session 4; telephone calls 'gradually reduced' after session 5	No	Yes	3 and 12 months
RCT comparisons of intervention delivery							
Beresford *et al.* (2012)[21]	10 weeks	Face to face	One (home)	Home visit approximately weekly for 6–8 weeks vs. telephone call approximately weekly for 6–8 weeks		No	10 and 22 weeks
Sciberras *et al.* (2011)[125]	1 session vs. 4 weeks	Face to face	One (clinic) vs. two (clinic)	None vs. telephone call ($n = 1$) followed by a face-to-face session (clinic) if needed	Yes, ADHD	No	2 months

NR, not reported.

The intervention was delivered in the home and/or clinic for four RCTs[107,124,125,138] and solely in the home for one.[21] All before-and-after study interventions were delivered in both homes and in clinics, apart from one study, in which the intervention was delivered solely in the home.[146] Implementation support in the RCTs, once regular sessions were completed, ranged from none,[125] to one or two further contacts[107,138] and to regular contacts over a period of several weeks.[21,124] All before-and-after studies had implementation support involving regular contact of between 6 and 28 weeks.[21,123,126,146]

The age of participants varied, with a mean age ranging from 2.67 to 12.1 years for the RCTs[21,107,124,125,138] and from 2.88 to 5.1 years for the before-and-after studies.[21,123,126,146] Most RCTs had more than one criterion for inclusion in the trial and included children with a mix of sleep disturbances (see *Table 6*) relating to sleep initiation only,[125,138] sleep initiation and maintenance[21,107] or sleep initiation, maintenance and scheduling.[124] All before-and-after studies included children with sleep initiation and maintenance disturbances.[21,123,126,146]

There was limited reporting of prior interventions. Two RCTs excluded children who were already receiving specialist sleep input[138] or who were using sleep supplements.[107] One RCT[124] reported that children were taking medication prescribed for behaviour and sleep problems; however, it was not clear if melatonin was being taken to aid sleep. One before-and-after study[126] reported that one child was taking medication for behaviour and sleep problems.

Twenty-seven different sleep-related outcomes were measured (see *Appendices 11–13*). First outcome measurement time points ranged from immediately post intervention to 2 months post intervention (see *Table 6*). Five trials measured additional follow-up time points; however, in this review we focus only on the outcomes closest to the end of the intervention.

Given the variation in interventions in terms of, for example, number of sessions, implementation support and mode of delivery, as well as differences in comparator and length of follow-up, a narrative synthesis was undertaken of the parent-directed tailored interventions.

Disorder global measures and composite scores

Total sleep time

Two RCTs ($n = 284$)[107,138] reported actigraphy-measured TST and a before-and-after study[126] presented parent-reported TST. However, because of the way in which data were reported, we are unable to calculate the MD from pre to post intervention or a CI.[126] Sleep management interventions seek to increase duration of TST.

There was no evidence of a statistically significant difference in change in actigraphy-measured TST from baseline to post intervention for a tailored parent-directed intervention compared with usual care (MD 10.9 minutes, 95% CI –19.0 to 40.8 minutes).[138] There was also no difference between the comparator and intervention groups post intervention for a group receiving a tailored parent-directed intervention compared with non-sleep-related parent education (MD 26.0 minutes, 95% CI –35.1 to 87.1 minutes).[107]

Sleep efficiency

Two RCTs ($n = 284$)[107,138] reported actigraphy-measured sleep efficiency, that is, the ratio of TST to total time in bed, informed by or verified using sleep diaries. Greater sleep efficiency is preferable. There was no evidence of a statistically significant difference in change in actigraphy-measured sleep efficiency from baseline to post intervention for a tailored parent-directed intervention compared with usual care (MD –1.6%, 95% CI –5.2% to 1.9%),[138] or in the difference between groups post intervention for a tailored parent-directed intervention compared with non-sleep-related parent education (MD –1.0%, 95% CI –7.6% to 5.6%).[107]

The Children's Sleep Habits Questionnaire

Four RCTs ($n = 310$)[21,124,125,138] and two before-and-after studies[21,123] reported the CSHQ total score. The CSHQ is a validated parent-reported assessment of child sleep.[133] Higher scores on the CSHQ indicate a

greater severity of the sleeping disturbance, either because of the frequency or number of different behaviours presenting.

There was a statistically significant reduction (i.e. improvement) in total CSHQ score in one RCT of a parent-directed tailored intervention compared with a usual care control. However, Hiscock *et al.*,[138] in another RCT, found no statistically significant improvement using a parent-directed tailored intervention compared with waiting list control,[124] although the direction of effect was in a positive direction. No evidence of a difference was observed in the RCTs of extended versus brief intervention[125] and face-to-face implementation support compared with telephone-delivered implementation support,[21] although the direction of effect was the same (*Table 7*).

There was a decrease (i.e. an improvement) in total CSHQ score post intervention compared with pre intervention in one before-and-after study;[123] however, it was not possible to estimate the MD between pre and post intervention in a second study as it was not a matched sample.[21]

TABLE 7 Outcome results for CSHQ score in parent-directed tailored interventions

Study	Time point, mean CSHQ score (SD)		MD (95% CI)
	Baseline	Follow-up	
RCTs			
Beresford *et al.* (2012)[21]	Intervention group: 59.50 points (11.82 points)	Intervention group: 52.17 points (11.44 points)	–1.16 points (–14.27 to 1.95 points)[a]
	Control group: 53.33 points (4.27 points)	Control group: 53.33 points (8.76 points)	
Hiscock *et al.* (2015)[138]	Intervention group: 57.8 points (8.8 points)	Intervention group: 50.1 points (8.3 points)	Adjusted: –6.6 points (–8.5 to –4.6 points)[b]
	Control group: 59.0 points (7.8 points)	Control group: 55.1 points (8.6 points)	–5.0 points (–7.6 to –2.4 points)[a]
Moss *et al.* (2014)[124]	Intervention group: 56.20 points (9.38 points)	Intervention group: 46.50 points (7.29 points)	–4.62 points (–10.83 to 1.59 points)[a]
	Control group: 51.38 points (7.54 points)	Control group: 51.12 (6.51)	
Sciberras *et al.* (2011)[125]	NR	Intervention group: (change score) 5.09 points (5.12 points)	–1.73 points (–7.11 to 3.65 points)[c]
		Control group: (change score) 6.82 points (8.02 points)	
Before-and-after studies			
Austin *et al.* (2013)[123]	55.43 points (7.68 points)	47.57 points (9.14 points)	–7.86 points (–14.39 to –1.33 points)[a]
Beresford *et al.* (2012)[21]	59.55 points (7.59 points)	56.57 points (10.77 points)	Cannot be estimated.[d] Effect size given as 0.42

NR, not reported.
a Estimated unadjusted difference at follow-up (post intervention) between intervention and control group for RCTs and change from pre to post intervention for before-and-after studies.
b Reported in paper.
c Difference in change scores from baseline to 2 months between intervention and control groups.
d As not a matched sample ($n = 11$ pre intervention and $n = 7$ post intervention).

Other global measures and composite scores

Several further composite scores and global outcome measures were reported by single studies. One RCT[138] ($n = 244$) of a tailored parent-directed intervention compared with usual care reported a statistically significant reduction in parent-reported moderate or severe sleep problems (*Table 8*). There appeared to be improvement at follow-up in three further outcomes measured in two before-and-after studies (see *Table 8*).[21,123]

Sleep initiation

Sleep onset latency

One RCT ($n = 40$)[107] reported actigraphy-measured SOL, the time from bedtime to sleep onset time, which was verified using sleep diaries. One before-and-after study reported on SOL using sleep diaries[123] and another reported SOL using parent-completed visual graphs.[126]

There was no evidence of a statistically significant difference in SOL in the RCT[107] of a tailored parent-directed intervention compared with non-sleep-related parent education (MD 4 minutes, 95% CI –15.0 to 23.0 minutes). One before-and-after study showed no statistically significant difference pre and post intervention (MD 43 minutes, 95% CI –30 to 116 minutes).[123] The results reported in the other study[126] are based on visual graphs without any associated numerical data; therefore, we are unable to present a MD and CI.

Other sleep initiation outcomes

Two further before-and-after studies measured time to settle at night. One reported a statistically significant improvement in time to settle at night (MD –91.3 minutes, 95% CI –112.6 to –69.9 minutes)[146] and the other did not report data for any sleep initiation outcomes.[126]

TABLE 8 Other global and composite outcomes of parent-directed tailored interventions

Study	Outcome	Time period, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
RCTs				
Hiscock *et al.* (2015)[138]	Moderate or severe sleep problems – parent/caregiver reported (none/mild and moderate/severe)	NR but eligibility criteria of 'parent reported moderate to severe sleep problems'	Intervention group: 30% Comparison group: 56%	Adjusted odds ratio 0.30 (0.16 to 0.59)[a] Difference in absolute risk 25.7% (14.1% to 37.3%)[a]
Before-and-after studies				
Austin *et al.* (2013)[123]	Sleep Disturbance Index	NR	NR	No data reported other than that pre- and post-treatment scores differed significantly ($z = -2.37$; $p < 0.05$)
Beresford *et al.* (2012)[21]	Parent-set child sleep goal(s)	2.64 (2.11)	6.50 (2.14)	Cannot be estimated[b]
Beresford *et al.* (2012)[21]	Change in goal attainment rating	N/A	Improved: 75.0% No change: 12.5% Deteriorated: 12.5%	N/A

NR, not reported; N/A, not applicable.
a Reported in paper.
b As not a matched sample ($n = 11$ pre intervention and $n = 8$ post intervention).

Sleep maintenance

Night wakings

Three before-and-after studies[123,126,146] reported night wakings using parent-reported diaries ($n = 46$), although each study defined the outcome differently.

One study reported a statistically significant reduction in the number of night wakings post intervention (MD –2.7, 95% CI –3.0 to –2.4).[146] Another showed a statistically significant reduction in the time in minutes it took to settle the child after night wakings (MD –24.0 minutes, 95% CI –44.7 to –3.3 minutes).[123] For the third study,[126] which measured number of night wakings per week, the results reported by the authors are based on visual graphs without associated numerical data; therefore, we are unable to present the MD and CI.

Other sleep maintenance outcomes

A variety of other outcomes related to sleep maintenance were reported by single studies. One RCT[138] ($n = 244$) reported WASO (night waking duration and/or frequency after the child falls asleep) and found no evidence of a statistically significant difference between a tailored parent-directed intervention and usual care (*Table 9*). Another RCT[125] ($n = 27$) comparing an extended versus brief intervention found that there was a greater reduction (i.e. now 'no' or only a 'mild' sleep problem) in sleep problems at 2 months for the extended intervention group compared with the brief intervention group. One before-and-after study[146] reported statistically significant improvements in mean minutes per night the child was awake, and number of nights the child does not sleep in their own bed. Another before-and-after study[126] reported measuring the number of nights per week that the child fell asleep in their own bed and the number of nights per week that the child co-slept; however, the authors reported insufficient data to determine the effects of the intervention on these outcomes (see *Table 9*).

TABLE 9 Other sleep maintenance outcomes of parent-directed tailored interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
RCTs: sleep maintenance				
Hiscock *et al.* (2015)[138]	WASO (minutes)	NR	NR	1.5 (–16.9 to 19.9)[a]
Sciberras *et al.* (2011)[125]	Parent/caregiver reported 'no' or 'mild' sleep problems	NR	Intervention group: 25% Control group: 64%	–39% (–74% to –4%)[b]
Before-and-after studies: sleep maintenance				
Quine and Wade (1991)[146]	Mean minutes per night the child was awake	70.2 (32.1)	3.2 (4.7)	–67.0 (–90.4 to –43.6)[c]
Quine and Wade (1991)[146]	Number of nights child does not sleep in own bed per week	5.6 (0.9)	0.1 (0.4)	–5.5 (–6.3 to –4.7)[c]
Weiskop *et al.* (2005)[126]	Number of nights per week that child fell asleep in own bed	NR	NR	NR
Weiskop *et al.* (2005)[126]	Number of nights per week that child co-slept	NR	NR	NR

NR, not reported.
a Reported in paper, difference in change from baseline to 3 months.
b Estimated unadjusted difference in percentage at 2 months.
c Estimated change from pre to post intervention for sample of 15 children with waking problems.

Child-related quality of life, daytime behaviour and cognition

Cognition outcomes

No studies reported child cognition outcomes.

Attention deficit hyperactivity disorder symptoms

Two RCTs ($n = 271$)[125,138] reported on the severity of ADHD symptoms using the total score of the ADHD Rating Scale IV,[165] which is a validated parent- or teacher-completed questionnaire that has been used to assess the presence and severity of symptoms by parents/carers of children with ADHD. A higher score indicates more severe problems.

The authors of a RCT[125] comparing an extended versus brief parent-directed tailored intervention reported only that there was 'minimal change in ADHD symptoms . . . across both groups at 2 and 5 months'.[125] There was a statistically significant reduction in symptom severity as measured by the parent-reported ADHD Rating Scale IV in another RCT[138] comparing a tailored parent-directed intervention with usual care (adjusted MD –3.7 points, 95% CI –6.1 to –1.2 points; unadjusted MD –3.5 points, 95% CI –6.5 to –0.5 points). However, the difference in the teacher-reported ADHD Rating Scale IV score was not statistically significant at 3 months' follow-up (adjusted MD –2.4 points, 95% CI –5.3 to 0.4 points; unadjusted MD –5.3 points, 95% CI –9.2 to –1.4 points).

Pediatric Quality of Life Inventory

Two RCTs[125,138] ($n = 271$) reported on the child's quality of life using the Pediatric Quality of Life Inventory (PedsQL) version 4.0. The PedsQL is a validated parent-completed questionnaire that assesses parents' perception of quality of life in paediatric patients with chronic health conditions. Higher scores indicate better health-related quality of life.

There was a statistically significant improvement in the PedsQL in one RCT[138] comparing a tailored parent-directed intervention with usual care (adjusted MD 9.4 points, 95% CI 5.6 to 13.2 points; unadjusted MD 10.6 points, 95% CI 6.0 to 15.2 points). In the other RCT, there was no statistically significant difference in change scores from baseline to 2 months between groups receiving the extended versus brief formats of a parent-directed tailored intervention on the PedsQL (MD 4.27 points, 95% CI –2.48 to 11.02 points).[125]

Daytime behaviour

Three RCTs ($n = 297$) of parent-directed tailored interventions reported other child-related quality of life, daytime behaviour and cognition outcomes.[124,125,138] These data are summarised in *Table 10*. Hiscock *et al.*[138] reported statistically significant improvements in the Daily Parent Rating on the Evening and Morning Behaviour Scale score, in parent- and teacher-reported Strengths and Difficulties Questionnaire scores and in backwards digit recall, a parent-directed tailored intervention, compared with the usual care control group.[138] Another RCT[124] reported that there was no statistically significant difference in the Developmental Behaviour Checklist when comparing a parent-directed tailored intervention with waiting list control. A third RCT[125] found that there was no statistically significant difference between extended and brief format sessions in the Daily Parent Rating on the Evening and Morning Behaviour Scale score.

For the before-and-after studies, one[123] reported a statistically significant improvement in the Developmental Behaviour Checklist[166] post intervention. A second before-and-after study[146] evaluated child daytime behaviour and cognition using the Behavior Problem Index (BPI)[167] and reported a statistically significant reduction in total BPI score. A third before-and-after study[126] evaluated children's sleep behaviour goals and reported improvements post intervention, although we were unable to report the MD and CI, as the authors provided insufficient data to enable this.

TABLE 10 Other outcomes of parent-directed tailored interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
Child-related quality of life, daytime behaviour and cognition				
RCTs				
Hiscock *et al.* (2015)[138]	Daily parent rating of evening and morning behaviour (total)	Intervention group: 22.6 points (5.0 points) Control group: 22.7 points (5.8 points)	Intervention group: 16.6 (5.8 points) Control group: 21.0 (5.8 points)	Adjusted: –4.7 points (–6.5 to –2.8 points)[a] –4.4 points (–6.2 to –2.6 points)[b]
Hiscock *et al.* (2015)[138]	SDQ – parent report (total)	Intervention group: 22.6 points (5.7 points) Control group: 21.9 points (5.4 points)	Intervention group: 18.6 points (5.0 points) Control group: 21.4 points (5.4 points)	Adjusted: –3.0 points (–4.3 to –1.7 points)[a] –2.8 points (–4.4 to –1.2 points)[b]
Hiscock *et al.* (2015)[138]	SDQ – teacher report (total)	Intervention group: 15.0 points (7.3 points) Control group: 17.2 points (6.8 points)	Intervention group: 13.5 points (6.6 points) Control group: 17.2 points (6.8 points)	Adjusted: –1.7 points (–3.4 to –0.1 points)[a] –3.7 points (–5.7 to –1.7 points)[b]
Hiscock *et al.* (2015)[138]	Backwards digit recall	NR	NR	Adjusted: 5.2 points (0.03 to 10.4 points)[a]
Moss *et al.* (2014)[124]	Developmental Behaviour Checklist (Parent Version)	Intervention group: 66.20 points (25.66 points) Control group: 72.29 points (16.50 points)	Intervention group: 57.70 points (27.10 points) Control group: 69.25 points (14.95 points)	11.55 points (–31.61 to 8.51 points)[b]
Sciberras *et al.* (2011)[125]	Daily Parent Rating on the Evening and Morning Behaviour scale	NR	Intervention group: (change score) 4.13 points (9.63 points) Control group: (change score) 0.67 points (3.77 points)	3.46 points (–2.43 to 9.35 points)[c]
Before-and-after studies				
Austin *et al.* (2013)[123]	Developmental Behaviour Checklist (Parent Version)	87.00 points (25.81 points)	74.57 points (27.96 points)	12.43 points (2.25 to 22.61 points)[d]
Quine and Wade (1991)[146]	BPI	13.0 points (4.6 points)	9.7 points (4.3 points)	–3.3 points (–5.1 to –1.5 points)[d]
Weiskop *et al.* (2005)[126]	Child's sleep behaviour goals	NR	12/25 goals (48%) were achieved with 100% success, and the mean achievement scale was 76.3%	N/A

BPI, Behavior Problem Index; N/A, not applicable; NR, not reported; SDQ, Strengths and Difficulties Questionnaire.
a Reported in paper.
b Estimated unadjusted difference at follow-up between intervention and control group.
c Estimated difference in change scores.
d Estimated change from pre to post intervention.

Other child-related sleep outcomes

Parent help-seeking for sleep problem

One RCT[138] ($n = 244$) collected parent reports of other professional help sought for their child's sleep. Parents of children in the intervention group were less likely than parents of children in the control group to seek help for their child's sleep from a health professional (e.g. general practitioner, paediatrician or psychologist) (14% vs. 20% at 3 months), but this difference was not statistically significant.

School attendance

Two RCTs ($n = 271$) reported on school attendance, defined as the number of days missed from school.[125,138] Both reported only that there was no difference in this outcome between the intervention groups.[125,138]

Parent-related outcomes

Three RCTs[124,125,138] and one before-and-after study[146] reported parent-carer-related outcomes. The outcomes measured and methods of assessment are summarised in *Appendix 12*.

Depression Anxiety Stress Scales

Two RCTs ($n = 271$) reported on parental stress using the Depression Anxiety Stress Scales (DASS).[125,138] The DASS are a validated self-report instrument designed to measure the three related negative emotional states of depression, anxiety and tension/stress. Higher scores indicate worse depression, anxiety and stress.

There was no evidence of a statistically significant difference in the DASS for a parent-directed tailored intervention compared with usual care (adjusted MD –5.3 points, 95% CI –13.0 to 2.4 points; unadjusted MD –5.3 points, 95% CI –13.1 to 2.5 points).[138] Sciberras *et al.*[125] found no evidence of a statistically significant difference in change scores from baseline to month 5 (data not available for 2-month follow-up) between the extended and brief intervention groups (MD 1.82 points, 95% CI –1.03 to 4.67 points).

Other parent-/carer-related outcomes of parent-directed tailored interventions

Two RCTs ($n = 53$)[124,125] and one before-and-after study[146] reported other parent-/carer-related outcomes of parent-directed tailored interventions. These data are summarised in *Table 11*. These outcomes focused on parental stress and morale[124,146] and parent work attendance.[125]

There was a statistically significant reduction (i.e. improvement) in the Parenting Stress Index – Short Form when comparing a parent-directed tailored intervention with a waiting list control.[124] Another RCT[125] found a statistically significant difference in parent work attendance for an extended versus a brief version of a parent-directed tailored intervention.

The before-and-after study found both a statistically significant reduction in maternal stress, as measured by the Malaise Inventory, and an improvement in maternal morale, as measured by the Cantrill ladder, from pre to post intervention.[146]

Measures of perceived parenting confidence and/or efficacy and/or understanding of sleep/sleep management

One RCT[21] and two before-and-after studies[21,146] reported on measures of perceived parenting confidence and/or efficacy and/or understanding of sleep/sleep management. Details of how these were measured are summarised in *Appendix 13*.

Parenting Sense of Competence scale and satisfaction and efficacy subscales

One RCT ($n = 13$)[21] and one before-and-after study ($n = 12$)[21] used the Parenting Sense of Competence (PSOC) scale.[168] The PSOC scale is a validated self-completed questionnaire. A higher score indicates a higher sense of parenting competence.

TABLE 11 Other parent-/carer-related outcomes of parent-directed tailored interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
RCTs				
Moss *et al.* (2014)[124]	Parenting Stress Index – Short Form	Intervention group: 89.30 points (18.70 points) Control group: 103.63 points (20.04 points)	Intervention group: 80.50 points (15.23 points) Control group: 103.00 points (21.84 points)	–22.50 points (–39.02 to –5.98 points)[a]
Sciberras *et al.* (2011)[125]	Parental work attendance	NR	NR	'At three months, intervention parents reported fewer days late for work as a result of their child's behaviour than control parents ($p = 0.02$, non-parametric test for trend) and fewer missed days at work ($p = 0.03$)'
Before-and-after studies				
Quine and Wade (1991)[146]	Maternal stress (Malaise Inventory)	6.4 points (4.1 points)	3.8 points (2.8 points)	–2.6 points (–1.5 to –3.7 points)[a]
Quine and Wade (1991)[146]	Maternal morale (Cantrill ladder)	6.7 points (2.2 points)	7.6 points (1.3 points)	0.9 points (0.2 to 1.6 points)[a]

NR, not reported.
a Estimated unadjusted difference at follow-up between intervention and control group for RCTs and change from pre to post intervention for before-and-after studies.

No evidence of a statistically significant difference was found in the RCT comparing modes of delivering implementation support (face to face vs. telephone) for either the satisfaction (MD 6.17 points, 95% CI –5.75 to 18.09 points) or the efficacy (MD –0.50 points, 95% CI –9.66 to 8.66 points) subscales of the PSOC scale.[21] Both satisfaction and efficacy subscale scores increased post intervention in the before-and-after study; however, a MD and CI was not calculated, as the summary data presented were not reported for a matched sample pre and post intervention.[21]

Other measures of perceived confidence and/or efficacy and/or understanding of sleep/sleep management

One before-and-after study[146] measured improvement in parental knowledge of behavioural principles using the Knowledge of Behavioural Principles as Applied to Children test.[169] The Knowledge of Behavioural Principles as Applied to Children test is a validated multiple choice instrument that assesses verbal understanding of basic behavioural principles. Higher scores indicate better knowledge scores. There was a significant improvement between pre and post intervention in the parents' knowledge scores (MD 2.2 points, 95% CI 1.7 to 2.7 points).

Summary

Nine studies evaluated parent-directed, comprehensive tailored interventions.[21,107,123–126,138,146] There were five RCTs,[21,107,124,125,138] three of which[107,124,138] were rated as having a high risk of bias and compared the intervention with usual care or other control, one[125] of which was rated as having a high risk of bias and compared brief with extended versions of an intervention and another of which[21] was rated as having a high risk of bias and compared different modes of delivering implementation support. There were also four before-and-after studies[21,123,126,146] that were rated as having a high or an unclear risk of bias. The mean age of participants ranged from 2.7 to 12.1 years. The majority of studies[21,123–126,138,146] included children with a mixed range of NDs, with three studies[107,125,146] including children with a single condition. The majority of studies[21,107,123,126,146] also included children with a mix of sleep disturbances, mainly sleep initiation and/or sleep maintenance problems.

Although all of the studies used face-to-face sessions with parents (at home or in a clinic) as the primary mode of delivery and were similar insofar as the content was classified as comprehensive, there was considerable variation in the duration of the intervention, the number of sessions delivered and the extent of implementation support when the sessions had finished, for example through follow-up telephone contact.

Overall, there was mixed evidence about the effects of parent-directed tailored interventions compared with usual care or other control, with limited evidence of benefit on more objective actigraphy child-related sleep outcomes and more evidence of benefit on parent-reported child outcomes and a few parent outcomes. Conclusions are hampered by the limited number of RCTs, the multitude of outcome measures and the risk of studies being underpowered to detect an effect. A single study[138] was rated as having a low risk of bias for all domains except for blinding, as the outcomes may have been influenced by the lack of blinded outcome assessment. This RCT[138] of a parent-directed comprehensive tailored intervention (with implementation support) compared with usual care, which provided the best available evidence identified, reported a small increase in TST in favour of the intervention, but the lower CI covered the possibility of worsening of TST (MD 10.9 minutes, 95% CI –19.0 to 40.8 minutes). A second RCT of a parent-directed comprehensive tailored intervention (with implementation support) compared with usual care had a similar finding (MD 26.0 minutes, 95% CI –35.1 to 87.1 minutes).[107] There was also no statistically significant improvement in other actigraphy-measured sleep outcomes with the intervention compared with control (sleep efficiency, WASO, SOL), measured by either or both studies. There was a statistically significant improvement in child sleep habits (CSHQ) and in parent-reported child sleep problems in one of these studies,[138] as well as child behaviour and quality of life (parent-reported ADHD Rating Scale IV, PedsQL, Parent Daily Reporting of Morning and Evening Behaviour). The third RCT[124] reported no statistically significant difference in child behaviour (Developmental Behaviour checklist). One RCT[138] reported no statistically significant improvement in parental stress (DASS) with the intervention compared with control, whereas another RCT[124] reported a statistically significant improvement (Parenting Stress Index – Short Form). The four before-and-after studies[21,123,126,146] showed improvement across several child-related and parent-related outcomes.

Based on a single RCT,[21] there was no statistically significant difference in the clinical effectiveness of home-delivered compared with telephone-based implementation support on child sleep outcomes (CSHQ) or parental outcomes (PSOC scale). Based on a single RCT,[125] there was a statistically significant improvement in parental work attendance and parent-reported child sleep problems with an extended versus brief intervention but no difference for multiple other child-related and parent-related outcomes.

Parent-directed non-tailored interventions

Eight studies evaluated parent-directed non-tailored interventions (*Table 12*): two two-armed parallel-group RCTs,[49,127] one three-armed parallel-group RCT[49] and five before-and-after studies.[129,149,151,162,163] Non-tailored parent-directed intervention involves the delivery of a standard 'training curriculum'. The curriculum may include opportunities for a parent to be supported to operationalise the material learnt for their child's sleep disturbance. Some implementation support may also be included.

The interventions varied in terms of the number of training sessions, the mode of delivery and the provision of implementation support. The number of sessions ranged from zero (leaflet provision only)[127] to two for the RCTs[49] and from one[149,163] to three and four sessions for the before-and-after studies.[129,151,162] The duration of the intervention was relatively short for RCTs, up to 2 weeks,[128] but was longer for the before-and-after studies, ranging from a single session[149,163] to 7 weeks.[151] Interventions also varied in terms of whether or not 'implementation support' was provided to parents. Some did not offer any implementation support ($n = 3$), some offered it during training sessions only ($n = 2$) and others also offered it for a time-limited period after all the sessions had been completed ($n = 3$). The curricula of four of the interventions included the opportunity within sessions for parents to operationalise learning for their child's specific sleep problem.

TABLE 12 Overview of non-tailored interventions and study design

Study	Total duration of intervention	Intervention details (active arms only)				Was the intervention described as having been developed for a specific ND?	Manual?	Length of follow-up, from baseline
		Mode of delivery	Number of sessions over which the curriculum was delivered	Was there an opportunity to operationalise curriculum content for the child's sleep problem?	Mode of delivering implementation support, and intensity, once curriculum was delivered: mode and intensity			
RCTs								
Adkins *et al.* (2012)[127]	N/A	Booklet	N/A	No	None	Yes, ASD	N/A	2 weeks
Before-and-after studies								
Beresford *et al.* (2012)[162]	5 weeks	Group	4	Yes	None (but included within curriculum for group session)	No	Yes	5, 17 and 29 weeks
Beresford *et al.* (2012)[163]	1 session	Teaching workshop	1	No	None	No	Yes	12 and 24 weeks
Bramble (1997)[149]	1 session	Face to face (clinic)	1	Minimal ('only minor individual tailoring')	Telephone calls on 3 consecutive days after session. Additional calls arranged if necessary	No	Yes	2 weeks, 4 months and 18 months
Reed *et al.* (2009)[129]	3 weeks	Group	3	Yes	None (but included within curriculum for group sessions)	Yes, ASD	Yes	7 weeks
Yu *et al.* (2015)[151]	7 weeks	Group, plus weekly telephone calls	3	Yes	Weekly for 4 weeks	Yes, ASD	Yes	3, 7 and 11 weeks
RCT comparisons of intervention delivery								
Montgomery *et al.* (2004)[49]	N/A vs. 1 session	Booklet vs. face to face	N/A vs. 1	No vs. no	None vs. none	No	Yes	6 weeks
Malow *et al.* (2014)[128]	2 weeks	Face to face vs. group	1 vs. 2	Yes	Weekly telephone call ($n = 2$) after sessions completed	Yes, ASD	Yes	1 month

N/A, not applicable.

The RCTs investigated interventions delivered both via written materials and through individual work (face to face and telephone calls). The mode of delivery was in booklet format for two RCTs[49,127] and was face to face for the third RCT, which was supplemented with two weekly telephone calls.[128] The mode of intervention delivery for the before-and-after studies was predominantly in group format, apart from in one study.[149] Implementation support was included in two before-and-after studies, which took the form of telephone calls for 3 days following the initial session for one study[149] and weekly for 4 weeks for the other.[151]

The comparators in the three trials were no intervention,[127] individual versus group delivery of an intervention[128] and, in a three-arm trial, written material versus face-to-face delivery of material versus no intervention control.[49]

For four interventions, study authors reported that the content of the intervention was informed by the nature of the target population, namely children with ASDs.

One RCT and three before-and-after studies included children with a range of NDs.[49,149,162,163] Two RCTs and two before-and-after studies included children with ASD only.[127–129,151]

The age of participants ranged from 2 to 5.9 years in the RCTs,[49,127,128] and from 4.8 to 8.9 years for the before-and-after studies.[129,149,151,162,163] Children in the RCTs had problems with sleep initiation[127,128] and problems with both sleep initiation and maintenance.[49] Children in the before-and-after studies had problems with both sleep initiation and maintenance, with the exception of one study.[162] Only one study[149] reported on whether or not the child had received any prior interventions.

Table 12 provides an overview of the studies evaluating the non-tailored parent training interventions.

The RCTs[49,127,128] reported on five outcomes and the before-and-after studies[129,149,151,162,163] reported on 10 outcomes. The most commonly reported outcome measures used were the CSHQ,[127–129,151,162,163] TST,[127,128,151] SOL,[127,128,149,151] WASO[127,128,151] and child daytime behaviour.[128,129,149,151] No studies measured child cognition outcomes.

Sixteen different sleep-related outcomes were measured (see *Appendices 14–16*). The first outcome measurement time points ranged from 2 weeks to 7 weeks from baseline (see *Table 12*). Four trials[149,151,162,163] measured additional follow-up time points; however, in this review, we focus only on the outcomes closest to the end of the intervention.

Given the variations in intervention characteristics (e.g. mode of delivery, number of sessions, availability of implementation support), study design and study objectives, it was not considered appropriate to pool the RCT data. Therefore, the studies are described in a narrative synthesis.

Child-related outcomes

The outcomes measured and methods of assessment are summarised in *Appendix 14*.

Global measures and composite scores

Total sleep time

Two RCTs ($n = 116$)[127,128] and one before-and-after study ($n = 25$)[129] evaluated actigraphy-measured TST informed by the use of sleep diaries following existing guidance on the interpretation of actigraphy data.[134]

In one of the RCTs,[127] no evidence of a statistically significant difference in actigraphy-measured TST was found for an intervention comprising the provision of written information compared with no intervention (MD 12.2 minutes, 95% CI –25.1 to 49.5 minutes). A similar finding was reported by the RCT that compared the modes of delivering a sleep management training curriculum to parents (i.e. face to face vs. group) [MD –7.2 minutes (favours group intervention), 95% CI –29.7 to 15.3 minutes].[128] The authors also

report the findings of a post hoc analysis, pre and post intervention, for the two groups combined, which are not described in this report.

The before-and-after study[129] narratively reports that there were no 'significant changes' in TST following a group intervention.

Sleep efficiency

Two RCTs ($n = 116$)[127,128] reported actigraphy-measured sleep efficiency (the percentage of time spent in bed asleep) informed or verified by sleep diaries. The authors of the RCT[127] that evaluated the provision of sleep management training via written material, compared with no intervention, report a statistically significant difference in the mean change (baseline to treatment) for the two groups ($p = 0.04$); however, we were able to calculate the unadjusted difference between the two groups only post intervention and this difference was not statistically significant (MD 2.7%, 95% CI 2.0% to 7.4%). Similarly, no significant difference in this outcome was reported by the RCT[128] that compared face-to-face delivery with group delivery of an intervention (MD –1.1%, 95% CI –3.6% to 1.4%). The authors also report the findings of a post hoc analysis for sleep efficiency, pre and post intervention, for the two groups combined, which are not described in this report.

The Children's Sleep Habits Questionnaire

One RCT ($n = 80$)[128] and four before-and-after studies ($n = 126$)[129,151,162,163] reported the CSHQ total score. The CSHQ is a validated parent-reported assessment of child sleep.[133] Higher scores on the CSHQ indicate a greater severity of the sleeping disturbance resulting from either the frequency or number of different behaviours presenting. The RCT reported the CSHQ results only for all participants in the two treatment groups (combining participants receiving group and individual education) and not for each group separately.

For the RCT,[128] there was no statistically significant difference in total CSHQ score post intervention between the group receiving face-to-face delivery and the group delivery arm (*Table 13*). In a post hoc analysis, the authors report merging the two arms and report combined before-and-after outcomes, which are not described here. Two before-and-after studies, one evaluating a three-session group-delivered intervention[129]

TABLE 13 Outcome results for CSHQ of parent-directed non-tailored interventions

Study	Time point, mean CSHQ score (SD)		MD (95% CI)
	Baseline	Follow-up	
RCTs			
Malow *et al.* (2014)[128]	NR	NR	NR
Before-and-after studies			
Beresford *et al.* (2012)[162]	57.86 (9.76)	51.79 (8.91)	Cannot be estimated.[a] Effect size given as 0.20
Beresford *et al.* (2012)[163]	56.58 (9.50)	55.56 (10.76)	Cannot be estimated.[b] Effect size given as 0.02
Reed *et al.* (2009)[129]	56.63 (9.21)	49.74 (9.24)	–6.89 points (–2.58 to –11.2 points)[c]
Yu and Hong (2015)[151]	55.11 (8.38)	51.76 (7.53)	–3.34 points (–1.4 to –5.3 points)[d]

NR, not reported.
a MD could not be calculated as it is not a matched sample (pre intervention, $n = 21$; post intervention, $n = 14$).
b MD could not be calculated as it is not a matched sample (pre intervention, $n = 24$; post intervention, $n = 18$).
c The change from pre to post intervention.
d The change from pre to post intervention, which was calculated using the SD of MD reported in the paper, from which the 95% CI was estimated.

and the other evaluating a four-session group-delivered intervention with implementation support,[151] report a statistically significant improvement (i.e. a decrease) in CSHQ total score post intervention compared with pre intervention. For two additional before-and-after studies,[162,163] it was not possible to estimate the MD in total CSHQ score between pre and post intervention, as they were not matched samples; however, the studies report small or very small effect sizes.

Parent-set child sleep goals

Two before-and-after studies ($n = 47$) – one evaluating a single-session group-delivered intervention[163] and the other evaluating a group-delivered intervention over four sessions[162] – reported parent-set child sleep goals using a goal attainment rating scale. This was a 10-point rating scale used to indicate the extent to which a goal has been achieved, from 1 = very far from this goal to 10 = I have achieved my goal.

Both studies reported that, post intervention, the mean goal attainment rating had 'significantly improved' compared with the pre-intervention rating. It was not possible to estimate the MD between the scores at pre and post intervention, as neither study reported data for a matched sample.

These same before-and-after studies[162,163] also reported the proportion of children that had improved, deteriorated or not changed in the goal attainment rating (improvement/deterioration based on whether or not the goal scores have moved 1 point in a positive or negative direction). Both studies reported that, post intervention, the majority of parents reported an improvement in goal attainment rating compared with pre intervention. In one study,[162] 93% of participants had improved and there was no change in the other 7% and, in the other study,[163] 65% improved, 19% had no change and 16% had deteriorated.[163]

Family Inventory of Sleep Habits

One RCT ($n = 80$)[128] and two before-and-after studies ($n = 79$)[129,151] evaluated child sleep outcomes using the Family Inventory of Sleep Habits (FISH). FISH is a validated parent-completed scale to assess sleep hygiene in children with ASD[170] and a higher score indicates better sleep hygiene.

The RCT[128] reported the results of a post hoc analysis for all participants in the two treatment groups (group and individual education) combined and not for each group separately; therefore, these data are not reported here. One of the before-and-after studies[129] also did not report data for the total score (only for each item separately) and, therefore, they are not reported here. The other before-and-after study[151] of a group-delivered sleep management intervention reported statistically significant improvements (data reported only at week 11) in total FISH score post intervention compared with pre intervention (MD 1.38, 95% CI 0.05 to 2.71).

Composite sleep disturbance score

One RCT ($n = 66$)[49] reported a composite sleep disturbance score. The composite sleep disturbance score was calculated from parent-completed diaries to identify sleep problems, covering settling frequency, settling duration, night waking frequency and night waking duration. A higher score on the composite sleep disturbance score indicates more severe sleep problems.

This study compared no intervention with either the intervention delivered in written format or via a face-to-face session and it reported that there was no statistically significant difference between these delivery methods post intervention (MD 0.15, 95% CI –1.38 to 1.68). The trial also included a third, 'control', arm that received no treatment for the first 6 weeks after randomisation, following which participants were re-randomised to receive either the conventional treatment or the booklet treatment. The authors compared the two active arms (written format and face to face) with the control and found statistically significant differences between the written format group and the control group (MD 3.20, 95% CI 1.89 to 4.51), and between the face-to-face format and the control group (MD 3.35, 95% CI 2.32 to 4.38).[49]

Severity of sleep problems

One before-and-after study, evaluating an individually delivered parent-directed tailored intervention, used a visual analogue scale to capture parent-reported 'severity of sleep problems', ranging in score from 0 = no problem to 10 = very severe problem.[149] The author reported a statistically significant improvement for the group post intervention compared with pre intervention but does not present sufficient data to allow for a MD and 95% CI to be calculated.

Sleep initiation

Sleep onset latency

Two RCTs ($n = 116$)[127,128] and two before-and-after studies ($n = 40$)[129,149] listed SOL (the time from bedtime to sleep onset time) as an outcome. Bramble[149] described SOL as 'mean time to settle', whereas the other studies described SOL as the time from bedtime to sleep onset time.

In the RCT evaluating the provision of written information, no evidence of a statistically significant difference in SOL was found between this intervention and no intervention (*Table 14*).[127] Similarly, the authors of the second RCT[128] comparing modes of delivery (individual vs. group) reported that they found no evidence of a statistically significant difference between the two groups in terms of this outcome. The authors also report the findings of a post hoc analysis for SOL, pre and post intervention, for the two groups combined, which are not described in this report.

One before-and-after study[129] only narratively reports that there were no 'significant changes' in SOL following a group intervention. The other before-and-after study[149] that evaluated 'mean time to settle' reported a statistically significant reduction in mean time to settle following a face-to-face session.

Sleep maintenance

Wake after sleep onset

The outcome measure was WASO (night waking duration and/or frequency after the child falls asleep) in two RCTs ($n = 116$)[127,128] and one before-and-after study.[129]

TABLE 14 Outcome results for SOL of parent-directed non-tailored interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
RCTs				
Adkins *et al.* (2012)[127]	SOL (minutes)	Intervention group: 56.7 (27.1)	Intervention group: 49.5 (26.7)	–11.8 (–37.3 to 13.7)
		Control group: 52.1 (25.1)	Control group: 61.3 (47.0)	
Malow *et al.* (2014)[128]	SOL (minutes)	Individual: 59.8 (31.6)	Individual: 39.5 (21.6)	–0.2 (–9.9 to 9.5)
		Group: 56.0 (25.2)	Group: 39.7 (21.5)	
Before-and-after studies				
Reed *et al.* (2009)[129]	SOL (minutes)	NR	NR	NR
Bramble (1997)[149]	Mean time to settle (minutes)	58.6 (24.6)	15.8 (7.8)	–42.8 (–61.0 to –24.6)

NR, not reported.

The RCT[127] comparing an intervention (provision of written material) with no intervention found that there was no evidence of a statistically significant difference at follow-up between the two groups (MD 0.5, 95% CI –18.9 to 19.9). The RCT[128] comparing individual and group delivery of an intervention also reported that there was no statistically significant difference between the groups on this outcome measure (MD –0.2, 95% CI –9.9 to 9.5). When the two treatment groups were combined, the mean change from pre to post intervention was –3.5 minutes (95% CI –7.3 to 0.3 minutes). The before-and-after study only narratively reports that there were no 'significant changes' in WASO following a group intervention.[129]

Child behaviour

Child behaviour was evaluated using four different outcome measures: (1) the Child Behavior Checklist (CBCL), (2) the Repetitive Behaviour Scale – Revised (RBS-R), (3) the Parental Concerns Questionnaire (PCQ) and (4) the BPI.

The Child Behavior Checklist

One RCT ($n = 80$)[128] and one before-and-after study ($n = 54$)[151] used the CBCL to investigate changes in daytime behaviour problems.[171] The CBCL is a validated parent-completed outcome measure for identifying problem behaviour in children, which is comprised of eight subscales. A higher score indicates more severe problems. Although the before-and-after study reported a total CBCL score, the RCT reported the following subscales of the CBCL only: anxious/depressed, attention, ADHD and withdrawn.

The RCT comparing individual with group delivery of an intervention reported that there was no difference between the two arms at follow-up, although data were not presented separately for the two groups.[128] With the two groups combined, the mean change from baseline to post intervention was as follows: anxious/depressed –2.2 (95% CI –3.7 to –0.7), attention –3.3 (95% CI –5.4 to –1.2), ADHD 0.54 (95% CI –1.3 to 2.4) and withdrawn –2.8 (95% CI –4.8 to –0.8).

The before-and-after study evaluated a group-delivered intervention (with implementation support offered via telephone) and did not find evidence of a statistically significant improvement in total CBCL scores post intervention (week 7) (MD –0.91, 95% CI –2.48 to 0.66).[151]

The Repetitive Behaviour Scale – Revised

One RCT ($n = 80$)[128] and one before-and-after study ($n = 25$)[129] evaluated child daytime behaviour using the RBS-R (total).[172] A lower RBS-R score indicates fewer problems.

The authors of the RCT, comparing group and individual delivery, reported that they found no evidence of a statistically significant difference in the change from baseline in daytime behaviours between the two groups post intervention (data not provided). The authors also report the findings of a post hoc analysis for RBS-R, pre and post intervention, for the two groups combined, which are not described in this report.[128] In the before-and-after study[129] that evaluated a group-delivered intervention, a statistically significant improvement (i.e. decrease) was observed post intervention compared with pre intervention (MD –1.22, 95% CI –2.06 to –0.38).

The Parental Concerns Questionnaire

Two before-and-after studies ($n = 79$)[129,151] evaluated child daytime behaviour using the PCQ.[173] The PCQ is designed as a parent interview screening instrument assessing the severity of developmental and associated psychiatric symptomatology using a four-point scale. A higher PCQ score indicates more severe problems.

Both of the before-and-after studies[129,151] evaluated group-delivered interventions, one of which included implementation support.[151] A total score could not be derived for one study,[129] as the authors did not report this (data reported for each subscale of the PCQ separately). The other study[151] found no evidence of a statistically significant change in the PCQ scores post intervention (data reported for week 11 only) compared with pre intervention (MD –1.38, 95% CI –2.82 to 0.06).

The Behavior Problem Index

One before-and-after study ($n = 15$)[149] evaluated child daytime behaviour using the BPI.[167] The BPI is a validated parent-reported outcome measure of child behaviour. Higher scores indicate a greater level of behaviour problems.

The authors reported the BPI at 4 months' follow-up only and reported a statistically significant improvement for the group post intervention, with a score of 22.1 (SD 12.4), compared with a pre-intervention score of 32.6 (SD 13.6) (MD –10.5, 95% CI 3.3 to 17.7).[149]

Child's health-related quality of life

One RCT ($n = 80$)[128] measured health-related quality of life using the PedsQL.[174] The PedsQL is a validated parent-completed questionnaire that assesses parents' perception of quality of life in paediatric patients with chronic health conditions. Higher scores indicate better health-related quality of life.

The authors of the RCT,[128] comparing group delivery with individual delivery of a sleep management intervention, reported that they found no evidence of a statistically significant difference in the change from baseline in PedsQL score between the two groups post intervention (data not provided). The authors also report the findings of a post hoc analysis for PedsQL, both pre and post intervention, for the two groups combined, which are not described in this report.

Parent outcomes

Several studies assessed parent outcomes. The outcome domains assessed were mental well-being,[129,149,151] quality of sleep[149,151] and parents' sense of competence.[128,162,163]

Quality of sleep

A before-and-after study ($n = 15$)[149] of an individually delivered intervention with implementation support measured self-reported quality of sleep using the Maternal Sleep Scale.[175] The Maternal Sleep Scale is a validated self-completed questionnaire that measures mothers' sleep quality, with higher scores indicating better sleep quality.

The authors reported the Maternal Sleep Scale scores at the end of the treatment phase and reported a statistically significant improvement for the group post intervention, with a mean score of 7.1 (SD 2.3), compared with a pre-intervention mean score of 4.1 (SD 2.3) (MD 3, 95% CI 1.7 to 4.3).[149]

A second before-and-after study ($n = 54$)[151] of a group-delivered intervention with implementation support used the Pittsburgh Sleep Quality Index to measure parents' self-reported quality of sleep. The Pittsburgh Sleep Quality Index is a validated self-report questionnaire that assesses sleep quality.[176] Lower scores denote a healthier sleep quality. No statistically significant change on this measure was reported following the intervention (week 7) (MD –0.51, 95% CI –1.37 to 0.35).[151]

Parent/carer mental well-being

Two before-and-after studies ($n = 79$)[129,151] assessed parental stress using the Parenting Stress Index – Short Form.[177] The Parenting Stress Index – Short Form is a validated self-completed outcome measure used for measuring parenting stress. A higher score indicates higher levels of parental stress.

Both studies evaluated group-delivered interventions, one with implementation support[151] and one without.[129] Neither reported a statistically significant change on the Parenting Stress Index – Short Form in post-intervention compared with pre-intervention scores (*Table 15*).

A further before-and-after study[149] assessed psychological distress using the Malaise Inventory. The Malaise Inventory is a validated self-completed questionnaire that measures psychological distress.[178] A higher score on the Malaise Inventory indicates higher psychological distress. This study evaluating an individually

TABLE 15 Outcome results for parent/carer mental well-being of parent-directed non-tailored interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
Reed *et al.* (2009)[129]	Parental Stress Index – Short Form	96.10 (23.40)	94.00 (23.00)	–2.1 (–12.9 to 8.7)
Yu and Hong (2012)[161]	Parental Stress Index – Short Form	82.55 (8.15)	81.73 (8.32)	0.82 (–1.63 to 3.27)
Bramble (1997)[149]	Malaise Inventory	8.7 (4.3)	4.7 (3.9)	–4.0 (–1.7 to –6.3)

delivered sleep intervention with implementation support reported a statistically significant improvement in scores on the Malaise Inventory post intervention compared with pre intervention (see *Table 15*).[149]

Parenting sense of competence

One RCT ($n = 80$)[128] and two before-and-after studies ($n = 47$)[162,163] assessed parents' sense of competence using the PSOC scale.[168] The PSOC scale is a validated self-completed questionnaire. A higher score indicates a higher parenting sense of competence. This scale comprises two subscales: sense of efficacy and parenting satisfaction.

The authors of the RCT,[128] comparing group and individual delivery, reported that they found no evidence of a statistically significant difference in the change from baseline in PSOC between the two groups post intervention (data not presented). The authors also reported the findings of a post hoc analysis for parenting sense of competence, both pre and post intervention, for the two groups combined, which are not described in this report.

One before-and-after study[162] reported a statistically significant improvement in the PSOC-efficacy (effect size 0.82), but not PSOC-satisfaction (effect size 0.38), from pre to post intervention, following a four-session group intervention; however, a MD and 95% CI could not be calculated, as data were not reported for a matched sample. The second before-and-after study[163] reported very little change in scores on PSOC-efficacy and PSOC-satisfaction 12 weeks after attending a single-session workshop (effect sizes –0.15 and 0.11, respectively); however, a MD and 95% CI could not be calculated as data were not reported for a matched sample.

Summary

Eight studies[49,127–129,149,151,162,163] evaluated parent-directed non-tailored interventions to manage sleep disturbance in children with NDs. We defined non-tailored interventions as those comprising the delivery of a standard 'training curriculum'. All the non-tailored interventions were comprehensive in their content, covering material on sleep and sleep processes, sleep hygiene and the management of specific problem behaviours (e.g. night wakings). Within this set of interventions, the design of the curriculum may include opportunities for parents to be supported to operationalise the material learnt for their child's sleep disturbance. Some implementation support (i.e. support to parents as they implement new ways of managing their child's sleep disturbance) may also be included.

Compared with the tailored interventions reported in the previous section, the non-tailored interventions evaluated by studies included in this review were much more diverse in terms of mode of delivery [i.e. written material, group delivery (single vs. multiple sessions) and one-to-one work]. In addition, they differed in terms of the extent to which they could accommodate the specific information and training needs parents may have in terms of their own child's condition and/or sleep problem. Furthermore, some included implementation support, but others did not. When implementation support was provided, it was always via telephone calls.

The eight studies included one RCT[127] that compared intervention (written material) and no intervention and five before-and-after studies.[129,149,151,162,163] Three[129,151,162] evaluated a group-delivered intervention comprising three or four sessions, one[163] evaluated a single-session workshop and one evaluated an

individually delivered intervention.[149] One RCT[49] compared two modes of delivering the same training curriculum (written material vs. face-to-face session) with no intervention. The final RCT[128] compared delivery of the same training curriculum via group and individual delivery. All the studies were rated as having a high risk of bias.

Four interventions[127–129,151] were developed specifically for children with ASDs, and only children with these conditions were recruited to the studies. In the remaining four studies,[49,149,162,163] the interventions were evaluated using samples of children with a range of NDs. The mean age of children recruited to the studies ranged from 2.0 to 8.9 years. The majority of studies reported that the children had problems with both sleep initiation and sleep maintenance.

The conclusions that can be drawn are limited, owing to the rating of the high risk of bias, the use of post-intervention follow-up time points and the risk of studies being underpowered to detect an effect. In addition, a large number of different outcome measures were used and these evaluated a range of outcome domains. We organise the findings in terms of mode of delivery and, when relevant, availability of implementation support.

A single trial evaluated the impact of an intervention comprising the provision of written information (and no implementation support) with no intervention.[127] There was no evidence of benefit at 2 weeks post intervention, as measured using actigraphy-collected sleep outcomes data (TST, Sleep Efficiency, SOL and WASO). Another three-armed trial[49] compared the provision of written information or the delivery of the same material via a single face-to-face session with a waiting list control group. Neither active arm included the provision of implementation support. Evidence of benefit at 6 weeks post intervention, based on a parent-report measure (composite sleep disturbance score), was reported for both modes of intervention delivery compared with the control group (written information vs. control, MD 3.20, 95% CI 1.89 to 4.51; face-to-face delivery vs. control, MD 3.35, 95% CI 2.32 to 4.38). The study was not powered to detect a difference between the two active arms. No other child outcomes, or parent outcomes, were assessed by these studies. There is, therefore, mixed and very limited evidence regarding the impact that the provision of written information with no implementation support has on child sleep outcomes only. Given the differences in outcome measures, differences in post-intervention follow-up time points and the high risk of bias for both studies, it is not possible to draw further conclusions regarding this sort of intervention. Furthermore, no conclusions can be drawn regarding the relative benefits of written versus face-to-face delivery of training on sleep management.

One before-and-after study[149] evaluated an intervention comprising a single face-to-face training session and implementation support available for up to 2 weeks after the session. The authors report significant improvements post intervention on non-standardised sleep outcome measures and a standardised measure of child behaviour (BPI) but data were not fully presented. Given the study limitations, no conclusions can be drawn from this evidence.

Two before-and-after studies evaluated multisession ($n = 3$ or 4) group-delivered interventions for which no implementation support was available after the group sessions were completed.[129,162] In one study, outcomes were reassessed immediately post intervention;[129] in the other study, this took place 1 month post intervention.[162] Both studies report improvements in child sleep outcomes measured using the same standardised parent-report measure (CSHQ) (for Beresford *et al.*,[162] the pre- and post-intervention samples were not matched and the reported effect size was 0.2; for Reed *et al.*,[129] the MD was –6.89, with a 95% CI of –2.58 to –11.2) and, for one study (Beresford *et al.*[162]), using achievement of parent-set sleep goals (progress towards goals, 93% of participants; no progress, 7% of participants). However, one of the studies[129] also used actigraphy to collect sleep outcomes data (TST, SOL, WASO). No benefits were reported on these outcomes. Reed *et al.*[129] also reported evidence of benefit in terms of parent-reported child behaviour (RBS-R). A further before-and-after study[151] also evaluated a multisession ($n = 3$) group-delivered intervention but also offered implementation support after the group sessions had finished via weekly telephone calls (duration unclear). This study also used the CSHQ to measure child sleep outcomes

and reported significant improvements immediately post intervention (MD –3.34, 95% CI –1.4 to –5.3). Child behaviour outcomes were also assessed (CBCL and PCQ) and no benefits were reported. Beresford *et al.*[162] also report significant improvements in self-reported parental efficacy but not parenting satisfaction (PSOC scale); however, pre- and post-intervention samples were not matched so a MD and CI could not be calculated. Reed *et al.*[129] and Yu and Hong[161] investigated parenting stress using the Parenting Stressing Index; neither study reported a change in this outcome between pre- and post-intervention time points. Yu and Hong[161] also report no effect on parent-reported sleep quality (Pittsburgh Sleep Quality Index). Taken together, there is some limited evidence of benefit for group-delivered interventions in terms of parent-reported child sleep outcomes. We would again, however, note that these studies were rated as having a high risk of bias. It is not possible to draw further conclusions with respect to other child, or parent, outcomes.

A RCT[128] compared delivery of the same training curriculum via a multisession ($n = 2$) group-delivered intervention and a single intervention; both arms included the same level of implementation support. No differences in outcomes (1 month post intervention) between the two modes of delivery were reported on actigraphy-derived sleep outcomes (TST, Sleep Efficiency, SOL and WASO) or parent-reported measures of child sleep (CSHQ and FISH), behaviour (CBCL and RBS-R) or quality of life (PedsQL). As this is a single study that was rated as having a high risk of bias, and it is not clear if it was adequately powered, no conclusions can be drawn.

The final mode of intervention delivery evaluated was a single workshop (5 hours) delivered to groups of up to 20 parents.[163] No implementation support was available. Parent-reported sleep outcome measures (CSHQ and attainment of parent-identified sleep goals) were readministered 3 months after the workshop. Benefits on these indicators were reported by the authors (PSOC-efficacy, effect size 0.82; PSOC-satisfaction, effect size 0.38). However, MD and CIs could not be calculated, as the data were not reported for a matched sample. Given that this mode of intervention delivery was evaluated by only a single study that was rated as having a high risk of bias, no further conclusions can be drawn.

Non-comprehensive parent-directed interventions

Two studies evaluated parent-directed sleep management interventions, which we classified as being non-comprehensive in their content, that is, the content of the training was focused on just one topic area related to sleep disturbance (whereas interventions that were classified as comprehensive addressed sleep and sleep processes, sleep hygiene and the management of specific problem behaviours, such as night waking).

One intervention, evaluated using a cluster RCT[152] with an attention control arm, comprised a tailored intervention that was restricted to training parents on behavioural principles of managing problem sleep behaviours, including functional analysis and behaviour management strategies. A single session with a practitioner, delivered in families' homes, was followed by telephone calls on at least a weekly basis to support the implementation of sleep management strategies. The total duration of implementation support was variable, continuing up to 3 months after the session with the practitioner. A description of the 'attention control' intervention was not provided. Although this appears to be a cluster trial, the description of the recruitment and randomisation processes is unclear and, so, for the purposes of this analysis, we have treated it as if it were an individually randomised trial. There were insufficient data to allow for any potential clustering and, in any case, the nature of the clustering was unclear.

A before-and-after study[155] evaluated the second non-comprehensive intervention. This intervention was restricted to training parents on principles of sleep hygiene only and was a single session delivered in a clinic. No implementation support was provided.

The mean age of participants in the RCT[152] was 9.49 years and the children had a mixed range of NDs. Neither the children's ages nor their NDs were reported by the before-and-after study.[155] Neither study reported whether or not the child/parent had received any prior interventions.

A total of 37 sleep-related outcomes were measured in the RCT[152] (see *Appendices 15–17*). Follow-ups were conducted post intervention at 'visit 4' (approximately 1 month post randomisation) and 'visit 6' (approximately 3 months post randomisation). Here, when data are available for both time points, we report data from visit 4 as, for the majority of the sample, contact with a sleep practitioner had ceased by this point.[152] A total of four sleep-related outcomes were measured in the before-and-after study[155] (see *Appendices 15* and *17*). Follow-ups were conducted after 6 weeks, at the end of the intervention.

Table 16 provides an overview of the studies evaluating the non-comprehensive parent-directed interventions.

Child sleep-related outcomes

Global measures and composite scores

Both the RCT[153] ($n = 30$) and the before-and-after study[155] ($n = 23$) reported global sleep outcomes and composite scores, although they used different measures (*Table 17*).

The RCT[152] of the intervention restricted to training on behaviour modification principles found a statistically significant difference in Composite Sleep Index-measured child sleep compared with no intervention at 'visit 4'. There was no statistically significant difference in actigraphy-measured TST for children in the intervention and control groups at visit 4. The before-and-after study,[155] evaluating the intervention providing training in sleep hygiene, reported a statistically significant improvement in CSHQ total score at 6 weeks post intervention.

Sleep maintenance

The RCT[152] ($n = 30$) of the intervention restricted to training on principles of behaviour modification reported a variety of actigraphy-measured sleep maintenance outcomes.

TABLE 16 Details of 'non-comprehensive' parent-directed interventions

Study	Total duration of intervention	Mode of delivery	Number of sessions and location	Opportunity to operationalise curriculum content for child's sleep problem?	Mode of delivering implementation support, and intensity, once regular sessions with practitioner were completed	Intervention described as developed for specific ND?	Manual?
RCTs							
Wiggs and Stores (1998)[152]	Unclear	Face to face (home)	1 (home)	Yes	Weekly telephone calls. Continued for at least 1 month, total duration unclear	No	Yes
Before-and-after studies							
Peppers, *et al.* (2016)[155]	1 session	Face to face (clinic)	1 (clinic)	Yes	None	Yes, ADHD	Unclear

TABLE 17 Outcome results for global measures and composite scores for other parent-directed interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
RCTs				
Wiggs and Stores (1998)[152]	Composite Sleep Index	Intervention group: 6.73 (2.31)	Intervention group: 3.79 (1.89)	–2.83 (–4.24 to –1.42)[a]
		Control group: 7.23 (2.26)	Control group: 6.62 (1.89)	
	TST (hours)	Intervention group: 9.2 (0.3)	Intervention group: 9.6 (0.6)	0.2 (–0.4 to 0.8)[a]
		Control group: 9.1 (0.9)	Control group: 9.4 (0.9)	
Before-and-after studies				
Peppers *et al.* (2016)[155]	CSHQ	50.13 (7.16)	43.74 (6.49)	6.4 (4.3 to 8.5)[b]

a The estimated unadjusted difference at follow-up between the intervention and control groups.
b As reported in the paper: the change from pre to post intervention.

There were statistically significant reductions in fragmentation index (percentage of immobile phases during sleep period that were ≤ 30 seconds of duration), movement during sleep and movement index (percentage of sleep period spent moving) for both groups from pre intervention to visit 4; however, the differences between the groups at follow-up were not statistically significant (*Table 18*).[152]

Other child outcomes

The RCT[152] ($n = 30$) comparing the intervention restricted to training on behaviour modification principles with an attentional-control-measured child daytime behaviour using the parent-completed, and teacher-completed, Aberrant Behavior Checklist (ABC) (*Table 19*).[179] No statistically significant differences on ABC subscales (parent, or teacher, report) between the intervention and control arms were found post intervention, except for the parent-reported stereotypies subscale.

TABLE 18 Outcome results for sleep maintenance of other parent-directed interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
Wiggs and Stores (1998)[152]	Fragmentation index (%)	Intervention group: 16.5 (3.7)	Intervention group: 11.9 (4.1)	0.0 (–3.7 to 3.7)[a]
		Control group: 17.8 (5)	Control group: 11.9 (5.7)	
	Movement during sleep	Intervention group: 1.9 (1.6)	Intervention group: 1.4 (0.5)	0.1 (–0.4 to 0.6)[a]
		Control group: 2.7 (2.9)	Control group: 1.3 (0.8)	
	Movement index (%)	Intervention group: 11.3 (5.1)	Intervention group: 9.0 (2.7)	0.2 (–1.9 to 2.3)[a]
		Control group: 13.1 (6.4)	Control group: 8.8 (3.0)	

a The estimated unadjusted difference at follow-up between the intervention and control groups.

TABLE 19 Other child outcomes for non-comprehensive parent-directed interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
RCTs				
Wiggs and Stores (1998)[152]	ABC (parent report)[a]			
	Inappropriate speech	Intervention group: 2.47 (2.88)	Intervention group: 2.36 (1.95)	–2.02 (–4.20 to 0.16)[c]
		Control group: 4.53 (5.13)	Control group: 4.38 (3.64)	
	Hyperactivity	Intervention group: 21.13 (9.69)	Intervention group: 18.07 (11.12)	–4.24 (–13.35 to 4.87)[c]
		Control group: 24.20 (14.18)	Control group: 22.31 (13.16)	
	Stereotypies	Intervention group: 3.93 (2.74)	Intervention group: 2.86 (2.44)	–3.60 (–5.95 to –1.25)[c]
		Control group: 6.07 (4.59)	Control group: 6.46 (3.71)	
	Lethargy	Intervention group: 7.20 (6.03)	Intervention group: 5.14 (7.10)	–2.55 (–8.52 to 3.42)[c]
		Control group: 8.13 (6.63)	Control group: 7.69 (8.78)	
	Irritability	Intervention group: 15.07 (7.40)	Intervention group: 12.07 (8.46)	–2.47 (–8.76 to 3.82)[c]
		Control group: 15.07 (7.79)	Control group: 14.54 (8.35)	
	ABC (teacher report)[b]			
	Inappropriate speech	Intervention group: 2.14 (3.51)	Intervention group: 1.08 (1.93)	–0.34 (–1.82 to 1.14)[c]
		Control group: 1.50 (1.95)	Control group: 1.42 (2.02)	
	Hyperactivity	Intervention group: 13.36 (8.53)	Intervention group: 10.75 (8.00)	2.42 (–4.53 to 9.37)[c]
		Control group: 14.43 (11.76)	Control group: 8.33 (10.43)	
	Stereotypies	Intervention group: 2.00 (2.39)	Intervention group: 1.50 (2.35)	–1.50 (–4.22 to 1.22)[c]
		Control group: 3.93 (5.88)	Control group: 3.00 (4.57)	
	Lethargy	Intervention group: 5.29 (4.12)	Intervention group: 4.00 (4.71)	–0.42 (–3.69 to 2.85)[c]
		Control group: 10.36 (10.34)	Control group: 4.42 (4.01)	
	Irritability	Intervention group: 7.93 (5.57)	Intervention group: 6.83 (6.83)	1.41 (–3.78 to 6.60)[c]
		Control group: 11.29 (11.19)	Control group: 5.42 (7.05)	

continued

TABLE 19 Other child outcomes for non-comprehensive parent-directed interventions (*continued*)

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
Before-and-after studies				
Peppers *et al.* (2016)[155]	Vanderbilt Assessment Scale – Parent Form (questions 1–9)	11.39 (7.75)	7.52 (8.41)	–3.87 (–7.37 to –0.37)[c]
	Vanderbilt Assessment Scale – Parent Form (questions 10–18)	9.30 (9.08)	6.39 (8.51)	–2.91 (–4.81 to –1.01)[c]

a Visit 4.
b Visit 6.
c The estimated unadjusted difference at follow-up between the intervention and control groups for RCTs and change from pre to post intervention for before-and-after studies.

The before-and-after study[155] ($n = 23$) of an intervention training in sleep hygiene principles reported a statistically significant improvement in scores on an assessment tool capturing severity of ADHD symptoms.

Parent outcomes

The RCT[152] ($n = 30$) comparing the intervention restricted to training on behaviour modification principles with an attentional control reported on a range of parent outcomes, including:

- maternal TST (actigraphy)
- maternal fragmentation index (actigraphy)
- maternal movement during sleep (actigraphy)
- maternal movement index (actigraphy)
- maternal daytime sleepiness (Epworth Sleepiness Scale)
- paternal daytime sleepiness (Epworth Sleepiness Scale)
- maternal satisfaction with their own sleep (six-point Likert scale)
- paternal satisfaction with their own sleep (six-point Likert scale)
- maternal stress (Malaise Inventory)
- paternal stress (Malaise Inventory)
- maternal perceived ability to control difficult sleep-related behaviour of their child (visual analogue scale)
- paternal perceived ability to control difficult sleep-related behaviour of their child (visual analogue scale)
- maternal perceptions of their partner's ability to control difficult sleep-related behaviour of their child (visual analogue scale)
- paternal perceptions of their partner's ability to control difficulty sleep-related behaviour of their child (visual analogue scale).

Further details of how these outcomes were measured are provided in *Appendix 22*. The data are summarised in *Table 20*.

We found no statistically significant differences between the groups at follow-up for any of these outcomes except father's satisfaction with child's sleep.

TABLE 20 Other parent outcomes for other parent-directed non-pharmacological interventions (Wiggs and Stores[152])

Outcome	Time period, mean (SD)		MD (95% CI)
	Baseline	Follow-up	
Global measures			
TST (maternal)	Intervention group: 6.9 (0.9)	Intervention group: 7.6 (0.7)	0.4 (–0.4 to 1.2)[a]
	Control group: 7.5 (0.8)	Control group: 7.2 (1.4)	
Sleep maintenance			
Fragmentation index (%)	Intervention group: 10.3 (4.5)	Intervention group: 9.7 (3.9)	–0.1 (–2.8 to 2.6)[a]
	Control group: 10.4 (3.9)	Control group: 9.8 (3.2)	
Movement during sleep	Intervention group: 1.6 (1.2)	Intervention group: 1.2 (0.5)	–0.4 (–1.3 to 1.9)[a]
	Control group: 1.5 (0.5)	Control group: 1.6 (1.7)	
Movement index (%)	Intervention group: 9.6 (3.7)	Intervention group: 7.6 (4.2)	0.4 (–2.2 to 3.0)[a]
	Control group: 10.6 (6.0)	Control group: 7.2 (2.4)	
Sleep scheduling (Epworth Sleepiness Scale)			
Mothers	Intervention group: 5.67 (4.32)	Intervention group: 4.07 (2.67)	–2.24 (–4.79 to 0.31)[a]
	Control group: 7.47 (4.94)	Control group: 6.31 (4.01)	
Fathers	Intervention group: 10.38 (5.03)	Intervention group: 9.83 (5.06)	0.08 (–3.26 to 3.42)[a]
	Control group: 8.00 (4.71)	Control group: 9.75 (3.77)	
Quality of sleep (satisfaction with own sleep)			
Mothers	Intervention group: 4.27 (1.16)	Intervention group: 2.93 (1.27)	–0.92 (–1.99 to 0.15)[a]
	Control group: 4.07 (1.44)	Control group: 3.85 (1.57)	
Fathers	Intervention group: 3.08 (1.26)	Intervention group: 2.17 (1.03)	–0.33 (–1.21 to 0.55)[a]
	Control group: 2.67 (1.30)	Control group: 2.50 (1.31)	
Other outcomes			
Maternal stress	Intervention group: 8.36 (4.27)	Intervention group: 6.14 (4.96)	–2.55 (–6.15 to 1.05)[a]
	Control group: 8.73 (3.51)	Control group: 8.69 (4.66)	
Paternal stress	Intervention group: 5.27 (2.61)	Intervention group: 4.25 (2.38)	–0.88 (–3.41 to 1.65)[a]
	Control group: 5.92 (3.85)	Control group: 5.13 (4.16)	
Satisfaction with child's sleep (mother)	Intervention group: 3.67 (1.53)	Intervention group: 3.29 (1.44)	0.02 (–1.00 to 1.04)[a]
	Control group: 3.53 (1.36)	Control group: 3.27 (1.27)	
Satisfaction with child's sleep (father)	Intervention group: 4.62 (1.19)	Intervention group: 2.50 (1.31)	–1.37 (–2.37 to -0.37)[a]
	Control group: 3.92 (0.90)	Control group: 3.87 (1.36)	
Satisfaction with ability to cope with child's sleep (mother)	Intervention group: 3.79 (1.53)	Intervention group: 2.43 (0.94)	–0.34 (–1.19 to 0.51)[a]
	Control group: 3.27 (1.10)	Control group: 2.77 (1.30)	
Satisfaction with ability to cope with child's sleep (father)	Intervention group: 3.83 (2.67)	Intervention group: 3.58 (1.19)	0.20 (–0.69 to 1.09)[a]
	Control group: 2.67 (1.44)	Control group: 3.38 (1.19)	

continued

TABLE 20 Other parent outcomes for other parent-directed non-pharmacological interventions (Wiggs and Stores[152]) (*continued*)

Outcome	Time period, mean (SD)		MD (95% CI)
	Baseline	Follow-up	
Perceived ability to control difficult sleep-related behaviour of own child (mother)	Intervention group: 5.51 (2.52) Control group: 4.87 (2.96)	Intervention group: 7.21 (2.15) Control group: 6.52 (2.00)	0.69 (–0.86 to 2.24)[a]
Perception of partner's ability to control difficult sleep-related behaviour of their child (mother)	Intervention group: 5.61 (3.00) Control group: 4.38 (2.42)	Intervention group: 5.05 (2.26) Control group: 6.19 (1.83)	–1.14 (–2.68 to 0.40)[a]
Perceived ability to control difficult sleep-related behaviour of own child (father)	Intervention group: 5.42 (3.26) Control group: 4.93 (2.96)	Intervention group: 5.35 (2.65) Control group: 5.34 (2.50)	0.01 (–1.92 to 1.94)[a]
Perception of partner's ability to control difficult sleep-related behaviour of their child (father)	Intervention group: 6.46 (2.53) Control group: 7.36 (2.26)	Intervention group: 5.60 (2.98) Control group: 6.35 (2.51)	–0.75 (–2.81 to 1.31)[a]

a The estimated unadjusted difference at follow-up between the intervention and control groups.

Measures of perceived confidence and/or efficacy and/or understanding of sleep/sleep management

Both the RCT[152] and the before-and-after study[155] reported on measures of perceived confidence and/or efficacy and/or understanding of sleep/sleep management.

The RCT[152] reported on parents' orientation to internal or external control beliefs as measured using the internality/externality control scale. These data are summarised in *Table 21* and details of how these outcomes were measured are provided in *Appendix 23*. There were significant between-group differences for the father measures. The before-and-after study[155] measured perceived confidence and efficacy using a parent satisfaction Likert-type scale. The authors narratively report that 87% of the responses were positive; however, it is unclear whether or not these positive responses related to child's sleep and/or the intervention.

Summary

Two studies evaluated tailored interventions that were non-comprehensive in terms of their content. Thus, although comprehensive parent-directed interventions (tailored and non-tailored) covered material on sleep and sleep processes, sleep hygiene and the management of specific problem behaviours (e.g. night wakings), non-comprehensive interventions focused on just one of these aspects. Both non-comprehensive interventions were, however, tailored interventions, that is, the intervention was personalised to the specific child and family context.

A RCT[152] evaluated a parent-directed intervention in which the content was restricted to behavioural principles of managing problem sleep behaviour. This intervention was delivered via a single face-to-face session followed by implementation support at least weekly for up to 3 months (duration as required). The comparator group was an attention control in which parents attended a single session with a practitioner (same duration as active arm, session content not described). We treated follow-up data collected 1 month after the face-to-face session as post-intervention outcomes. The sample included a range of NDs and the mean age of the children was 8.21 years. The rating of risk of bias was high.

Post intervention, an actigraphy-derived measure of sleep outcome (TST) was no different between the intervention and control arms, with similar findings for other actigraphy-derived measures (fragmentation index, movement during sleep and movement index). However, significant differences in scores post

TABLE 21 Measures of perceived confidence and/or efficacy and/or understanding of sleep/sleep management: non-comprehensive parent-directed interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
RCTs				
Wiggs and Stores (1998)[152]	Internality (mother)	Intervention group: 10.71 (3.29)	Intervention group: 11.71 (4.12)	1.86 (–1.04 to 4.76)[a]
		Control group: 9.60 (4.85)	Control group: 9.85 (3.63)	
	Externality (mother)	Intervention group: 12.29 (3.29)	Intervention group: 11.50 (4.05)	–1.58 (–4.46 to 1.30)[a]
		Control group: 12.93 (4.71)	Control group: 13.08 (3.64)	
	Internality (father)	Intervention group: 12.73 (5.14)	Intervention group: 13.64 (4.59)	3.26 (0.37 to 6.15)[a]
		Control group: 12.00 (4.77)	Control group: 10.38 (2.97)	
	Externality (father)	Intervention group: 10.27 (5.14)	Intervention group: 9.36 (4.59)	–3.27 (–6.16 to –0.38)[a]
		Control group: 11.00 (4.77)	Control group: 12.63 (2.97)	
Before-and-after studies				
Peppers *et al.* (2016)[155]	Parent satisfaction	NR	NR	*87% of the responses were positive*

NR, not reported.
a The estimated unadjusted difference at follow-up between the intervention and control groups.

intervention on a parent-reported child sleep measure (Composite Sleep Index), favouring the intervention group, were found (MD –2.83, 95% CI –4.24 to –1.42). In addition, post intervention, fathers' (but not mothers') ratings of satisfaction with the child's sleep (non-standardised measure) were higher for the intervention group than for the control group (MD –1.37, 95% CI –2.37 to 0.37).

The study also assessed child behaviour (parent and teacher report) using five subscales (inappropriate speech, hyperactivity, stereotypies, lethargy and irritability) of the ABC. Post intervention, no significant differences between the intervention and control arms were found on any subscale scores except parent-reported stereotypies. Here, a benefit to the intervention group was reported (MD –3.6, 95% CI –5.95 to 1.25).

In terms of mothers' and fathers' outcomes, sleep outcomes [actigraphy-derived (mothers only): TST, fragmentation index, movement during sleep; self-report: Epworth Sleepiness Scale, non-standardised measure of satisfaction with sleep] and other outcomes (psychological distress (Malaise Inventory), non-standardised measures of perceptions of own and partner's ability to control child's difficult sleep-related behaviour) were monitored. No differences between intervention and control arms were found post intervention for any of these outcomes.

Overall, given that there is just one trial – and it has a high risk of bias – no conclusions can be drawn regarding the effectiveness of parent-directed sleep management interventions in which the content was restricted to behavioural principles of managing problem sleep behaviour.

In the second non-comprehensive parent-directed intervention, the content was restricted to sleep hygiene.[155] This was evaluated using a before-and-after study design. The intervention, specific to children with ADHD, was delivered via a single session between parent and practitioner. No implementation support was offered. The ages of children recruited to the study were not reported. Risk of bias was high. Follow-up was 6 weeks post intervention.

Post intervention, scores on the parent-reported CSHQ had improved (MD 6.4, 95% CI 4.3 to 8.5). A measure of ADHD symptom severity was also used (Vanderbilt Assessment Scale), and improvements were also reported on this measure post intervention (Vanderbilt Assessment Scale questions 1–9, MD –3.87, 95% CI –7.37 to 0.37; Vanderbilt Assessment Scale questions 10–18, MD –2.91, 95% CI –4.81 to 1.01). (Changes in scores on a non-standardised measure of parent satisfaction were also reported, although it is unclear if this is capturing satisfaction with the child's sleep and/or the intervention per se).

In terms of this intervention, given that the evidence is limited to a before-and-after study with a high risk of bias, no conclusions regarding its impact on children's sleep or ADHD symptoms can be drawn.

Other non-pharmacological interventions

Seven studies[36,156–161] evaluated other types of non-pharmacological interventions (see *Table 9* and *Appendix 18*).

One crossover RCT compared valerian (dried and crushed whole root from the *Valeriana edulis* plant) with placebo;[156] one crossover RCT compared weighted blankets with placebo blankets;[36] one parallel RCT compared faded bedtime with response costs and bedtime scheduling;[157] three uncontrolled before-and-after studies evaluated light therapy and a behavioural programme,[158] an aquatic exercise programme[159] and acupuncture and ear-point taping;[161] and one controlled before-and-after study compared essential fatty acid supplements with placebo.[160]

The total intervention duration ranged from 12 days to 36 weeks[36,156,160,161] and was not specified in two studies.[158,159]

Three studies included children with a mixed range of NDs.[36,156,157] Two included children with learning disabilities,[158,161] one included children with a diagnosis of ASD[159] and one included children with a diagnosis of ADHD.[160]

The age of participants varied, with a mean age range of 6.7–10.8 years for the RCTs[36,156,157] and a mean age range of 2.9–8.8 years for the uncontrolled before-and-after studies.[158,159,161] The controlled before-and-after study reported the participants' ages as between 9 and 12 years.[160] All RCTs included children with sleep initiation and sleep maintenance disturbances.[36,156,157] The before-and-after studies included children with a mix of sleep disturbances (see *Table 9*) relating to sleep maintenance and 'lack of sleep consolidation',[158] sleep dysfunction,[159] 'sleep deprived'[160] and sleep initiation, maintenance and abnormal sleep state.[161]

There was limited reporting of prior interventions. One parallel RCT reported that children were excluded if they were receiving pharmacological interventions for sleep.[157] One before-and-after study reported that over half of the study sample had attended sleep clinics and centres for treatment, with some children receiving behavioural treatments.[158]

A total of 17 child sleep-related outcomes were measured by the RCTs (see *Appendix 18*). Commonly measured child sleep-related outcomes included SOL,[36,156,157] night waking,[36,156,157] TST,[36,156] sleep quality[36,156] and child daytime behaviour and cognition.[36,156] A total of 10 child sleep-related outcomes were measured by the before-and-after studies (see *Appendix 18*) and these were mainly measured by

single studies. The majority of outcomes reported by before-and-after studies were measured by single studies. However, TST was measured by two studies.[158,159]

In all studies, follow-up was conducted immediately following the completion of the intervention, and this varied between studies. For the RCTs, the follow-up times were at 2 weeks,[156] at 4 weeks[36] and after the last 10 days of an 'on average 8-week' treatment.[157] For the before-and-after studies, the follow-up times were at 10 weeks[160] and at 6 months.[158] For one before-and-after study, the length of the follow-up was unclear and it was reported as 'after treatment'.[161] The final study, Oriel *et al.*,[159] adopted an A–B–A withdrawal design, in which A was a control phase and B was a treatment phase. Outcomes were reported at 4 (A1), 8 (B) and 12 (A2) weeks from the start of the intervention.[159] We report all outcomes for Oriel *et al.*[159] at 8 weeks, which was when the intervention ceased.

Global measures and composite scores

Total sleep time

Two RCTs ($n = 78$)[36,156] and two before-and-after studies[158,159] ($n = 22$) reported TST (*Table 22*).

TABLE 22 Outcome results for TST of other non-pharmacological interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
RCTs				
Francis and Dempster (2002)[156]	TST (hours)	Combined: 9.93 (0.56)	Intervention group: 10.28 (0.43) Control group: 9.94 (0.7)	0.34 (–0.42 to 1.10)[a]
Gringras *et al.* (2014)[36]	TST (minutes) (actigraphy)	Combined: 452.8 (59.7)	Intervention group: 452.8 (65.0) Control group: 455.4 (65.8)	–4.2 (–13.6 to 5.2)[a]
	TST (minutes) (sleep diary)	Combined: 531.8 (109.6)	Intervention group: 528.9 (127.1) Control group: 513.0 (154.1)	15.9 (–6.8 to 38.6)[a]
Before-and-after studies				
Guilleminault *et al.* (1993)[158]	TST (minutes)	Responders: 204 (24) Non-responders: 192 (51) Overall: 196 (40)	Responders: 425 (55) Non-responders: 202 (40) Overall: 281 (48)	Responders: 221 (162 to 280)[b] Non-responders: 10 (–26 to 46)[b] Overall: 85 (59 to 111)[b]
Oriel *et al.* (2016)[159]	TST (minutes)	493.59 (60.51)	576.91 (37.48)	83.32 (17.55 to 149.09)[c]

a Estimated unadjusted difference between intervention and control groups at follow-up.
b Estimated change from pre to post intervention.
c Estimated change from A1 to B.

One study reported TST using actigraphy-verified sleep diary data,[156] one study measured TST using sleep diary and actigraph data;[36] and one measured TST via 'semiweekly' telephone calls from researchers to parents/guardians. Results show that there was no statistically significant difference in TST between valerian and placebo groups.[156] There was no statistically significant difference in actigraphy- or sleep diary-reported TST for weighted blankets compared with control blankets.[36] There was a statistically significant increase in TST between periods A1 and B following an aquatic exercise intervention[159] and from pre to post intervention following light therapy and a behavioural programme.[158]

Other global measures and composite scores

One RCT[36] ($n = 73$) and two before-and-after studies[161] ($n = 38$) reported further global measures and composite scores (*Table 23*). There was a statistically significant difference in Composite Sleep Disturbance Index score (but not sleep efficiency) with weighted blankets compared with placebo blanket.[36]

Two before-and-after studies[159,161] measured CSHQ total score (validated parent-reported assessment of child sleep, with higher scores indicating a greater severity of the sleeping disturbance),[159,161] although one reported this only for baseline.[159] A statistically significant reduction in total CSHQ score was reported following acupuncture and ear-point taping post intervention.[161]

Sleep initiation

Bedtime settling

Two RCTs[36,156] ($n = 78$) and one before-and-after study[159] ($n = 8$) reported SOL (*Table 24*). One RCT reported SOL using a sleep diary only[156] and one used a parental diary and actigraphy.[36] The before-and-after study measured SOL via telephone calls from parents to researchers.[159]

There was no statistically significant difference in SOL with valerian treatment compared with placebo[156] or following an aquatic exercise intervention.[159] There was no statistically significant difference in actigraphy- or parent-reported SOL for the RCT of weighted blankets.[36]

One RCT narratively reported exactly what time the child fell asleep at night.[157] Among the eight participants who had issues with sleep initiation at baseline, trained observers reported that three out of the four participants who received the faded bedtime and response costs intervention experienced improved sleep initiation during the last 10 days of treatment, with one participant continuing to have issues falling asleep.

TABLE 23 Other global measure and composite score outcomes of other non-pharmacological interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
RCTs				
Gringras *et al.* (2014)[36]	Sleep efficiency (%)	Combined: 72.7 (8.8)	Intervention group: 73.6 (9.5) Control group: 74.2 (8.0)	–0.3 (–1.7 to 1.1)[a]
	CSDI	Combined: 12.2 (2.1)	Intervention group: 10.8 (2.3) Control group: 11.4 (2.0)	–0.7 (–1.3 to –0.1)[a]
Before-and-after studies				
Yu and Hong (2012)[161]	CSHQ (total)	57.97 (4.58)	46.47 (5.13)	–11.50 (–13.33 to –9.67)[a]

CSDI, Composite Sleep Disturbance Index.
a Estimated unadjusted difference between intervention and control group at follow-up for RCTs and change from pre to post intervention for before-and-after studies.

TABLE 24 Outcome results for sleep initiation of other non-pharmacological interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
RCTs				
Francis and Dempster (2002)[156]	SOL (minutes)	Combined: 41.14 (20.99)	Intervention group: 23.49 (13.42) Control group: 39.14 (34.68)	–15.65 (–53.31 to 22.01)[a]
Gringras *et al.* (2014)[36]	SOL (minutes) (actigraphy)	Combined: 76.5 (46.1)	Intervention group: 71.4 (48.2) Control group: 70.6 (44.3)	2.1 (–5.5 to 9.7)[a]
	SOL (minutes) (sleep diary)	Combined: 69.9 (47.6)	Intervention group: 55.6 (37.8) Control group: 57.2 (42.8)	–1.6 (–6.7 to 3.5)[a]
Before-and-after studies				
Oriel *et al.* (2016)[159]	SOL (minutes)	38.95 (21.19)	21.76 (15.94)	19.11 (–40.95 to 6.57)[a]

a Estimated unadjusted difference between intervention and control group at follow-up for RCTs and change from pre to post intervention for before-and-after studies.

Of the three participants who reported sleep initiation issues at baseline and received 'bedtime scheduling' (the control), two participants improved, one worsened and one saw little difference to their sleep initiation after 10 days of treatment.[157]

Sleep maintenance

Night waking

Three RCTs[36,156,157] ($n = 92$ participants randomised) and one before-and-after study[159] ($n = 8$) reported night waking using actigraphy,[156] sleep diary,[36] observations[157] and telephone calls from parents to researchers.[159]

Results are summarised in *Table 25*. These show that there was no statistically significant reduction in time awake during the night (night waking) following valerian treatment.[156] There was no statistically significant difference in number of night wakings following an aquatic exercise programme,[159] although the baseline number of wakings was very low, or for weighted blankets compared with placebo blankets.[36]

Piazza *et al.*[157] narratively reported that all five participants who received the faded bedtime and response costs intervention saw improvements to night wakings. Following bedtime scheduling of the six control participants who had issues with night wakings at baseline, two participants improved, two saw little difference and one had worsened night wakings.[157]

Other sleep maintenance outcomes

Two RCTs[36,157] ($n = 87$) and one before-and-after study[158] ($n = 14$) reported other outcomes related to sleep maintenance (*Table 26*). The before-and-after study evaluating light therapy and a behavioural programme reported sleep diary-measured 'longest wake and sleep periods during a 24-hour cycle'. Data for this outcome were reported only graphically and it was not possible to estimate results from the graph.[158]

One RCT narratively reported that all six of the participants who received the faded bedtime and response costs intervention and had problems with early waking at baseline had improved on this outcome during

TABLE 25 Outcome results for night waking of other non-pharmacological interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
RCTs				
Francis and Dempster (2002)[156]	Nocturnal time awake (minutes)	Combined: 17.92 (8.21)	Intervention group: 6.85 (7.16) Control group: 8.38 (9.07)	–1.53 (–11.83 to 8.77)[a]
Gringras *et al.* (2014)[36]	Number of night wakings	Combined: 20.9 (8.0)	Intervention group: 19.5 (7.0) Control group: 19.5 (6.8)	–0.2 (–1.1 to 0.7)[a]
Before-and-after studies				
Oriel *et al.* (2016)[159]	Number of night wakings	0.99 (0.77)	0.37 (0.41)	–0.62 (–1.45 to 0.21)[a]

a Estimated unadjusted difference between the intervention and control groups at follow-up for RCTs and change from pre to post intervention for before-and-after studies.

TABLE 26 Other sleep maintenance outcomes for other non-pharmacological interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
RCTs				
Gringras *et al.* (2014)[36]	Time awake after sleep onset (minutes)	Combined: 85.4 (45.1)	Intervention group: 84.6 (42.6) Control group: 84.5 (41.5)	–2.5 (–9.5 to 4.5)[a]
	Proportion of nights with ≥ 1 waking	Combined: 0.3 (0.3)	Intervention group: 0.2 (0.3) Control group: 0.2 (0.3)	–0.01 (–0.06 to 0.04)[a]
Piazza *et al.* (1997)[157]	Hours of disturbed sleep	Intervention group: 1.44 (SD NR) Control group: 1.37 (SD NR)	Intervention group: 0.53 (SD NR) Control group: 1.10 (SD NR)	0.57[b]

NR, not reported.
a Estimated unadjusted difference between intervention and control group at follow-up.
b Data too limited to calculate 95% CI.

the last 10 days of treatment. In the control group, two out of the six participants reported improvements in early wakings, one worsened and three saw little or no change following bedtime scheduling.[157]

There was no difference between the treatment groups at follow-up in Gringas *et al.*[36] in terms of time awake after sleep onset and the proportion of nights with one or more awakening.

Sleep quality

Two RCTs[36,156] ($n = 78$) and one before-and-after study[160] ($n = 78$) reported quality of sleep (*Table 27*).

There was a statistically significant improvement in sleep diary-measured sleep quality from baseline for children who received both valerian treatment and placebo; however, the difference in this outcome at follow-up between the groups was not statistically significant.[156] There was also improvement in sleep quality for children who received fatty acids and placebo, as measured on a 'short questionnaire'.[160]

TABLE 27 Sleep quality outcomes of other non-pharmacological interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
RCTs				
Francis and Dempster (2002)[156]	Sleep quality	Combined: 5.34 (1.49)	Intervention group: 7.54 (1.47) Control group: 6.71 (1.29)	0.83 (–0.90 to 2.56)[a]
Gringras *et al.* (2014)[36]	Children's perceptions of sleep quality[b]	NR	Intervention group: smiley face 56%, neutral face 30% and unhappy face 14% Control group: smiley face 35%, neutral face 47% and unhappy face 18%	–
	Parent's perceptions of child's sleep quality[c]	–	Intervention group: more agitated, 0%; no different from usual, 44%; calmer, 35%; and N/A as few awakenings, 21% Control group: more agitated, 5%; no different from usual, 62%; calmer, 14%; N/A as few awakenings, 20%	–
	Sleep improvement (parent perceptions)[d]	NR	Intervention group: very much improved, 15%; much improved, 36%; minimally improved, 31%; no change, 13%; minimally worse, 3%; much worse, 1%; very much worse, 0% Control group: very much improved, 1%; much improved, 15%; minimally improved, 24%; no change, 49%; minimally worse, 6%; much worse, 4%; very much worse, 0%	–
Before-and-after studies				
Yehuda *et al.* (2011)[160]	Quality of sleep	ADHD-FA: 1.0 (0.8) ADHD-vehicle: 1.2 (0.7)	ADHD-FA: 3.8 (0.7) ADHD-vehicle: 1.4 (0.8)	ADHD FA: 2.8 (2.6 to 3.0) ADHD vehicle: 0.2 (–0.05 to 0.4)[e]

ADHD-FA, attention deficit hyperactivity disorder – fatty acid; N/A, not applicable; NR, not reported.
a Estimated unadjusted difference between the intervention and control groups at follow-up.
b 'Which best describes how you have felt about your sleep over the past 2 weeks?'.
c 'Compared with before the trial when my child was not using any special sensory blanket, if my child woke at night, he or she seemed . . .'.
d 'Compared with before the trial when child was not using any special sensory blanket, my child's sleep is . . .'.
e Change from pre to post intervention, reported separately for the two groups.

Gringras *et al.*[36] measured children's perceptions of sleep quality using a smiley face rating scale. Parents' perceptions of their child's sleep quality were also measured. Parents were asked 'Compared with before the trial, when my child was not using any special sensory blanket, if my child woke at night, he or she seemed: more agitated; no different from usual; calmer; and not applicable as so few awakenings'. Gringras *et al.*[36] also reported parent's perceptions of their child's sleep improvement. These data are summarised in *Table 27*.

One before-and-after study[158] evaluating light therapy plus a behavioural programme reported sleep diary-measured distribution of sleep bouts during a 24-hour cycle. Data for this outcome were reported graphically, with no narrative discussion from the authors, and could not be extracted.

Child-related quality of life, daytime behaviour and cognition

Two RCTs[36,156] ($n = 78$) reported on child daytime behaviour using a sleep diary[156] and the ABC.[36]

There was no statistically significant difference in daytime behaviour and cognition for total ABC score or subscale scores between the weighted blanket group and the control blanket group at 4 weeks from baseline (MD –2.3, 95% CI –5.4 to 0.8).[36] Francis and Dempster[156] used the sleep diary data to narratively describe anecdotal changes in child behaviour, but quantitative data were not reported.

Other outcomes

One RCT ($n = 73$) and one controlled before-and-after study ($n = 78$) reported other outcomes of interest.[36,160] The RCT reported that there were no significant differences for the total Sensory Behaviour Questionnaire or its subscales between the weighted blanket group and the control blanket group.[36] The controlled before-and-after study that compared essential fatty acid supplements with placebo reported a variety of other child-related outcomes at the end of the 10-week intervention period.[160] These data are summarised in *Table 28*.

Adverse events

Two out of the seven studies reported adverse events (*Table 29*).[36,160] One study[36] reported how adverse events were measured, namely through parents reporting to a 24-hour telephone number and weekly face-to-face or telephone-based reviews with parents. Five studies did not report adverse events.[156–159,161]

TABLE 28 Other outcomes of other non-pharmacological interventions

Study	Outcome	Time point, mean (SD)		MD (95% CI)
		Baseline	Follow-up	
Child-related quality of life				
Gringras *et al.* (2014)[36]	Sensory Behaviour Questionnaire	Combined: 148.8 (42.4)	Intervention group: 138.6 (41.3) Control group: 142.4 (46.2)	–4.9 (–10.2 to 0.4)[a]
Other outcomes				
Yehuda *et al.* (2011)[160]	Degree of fatigue in general during the day	ADHD-FA: 2.3 (1.0)	ADHD-FA: 4.0 (0.3)	ADHD-FA: 1.7 (1.4 to 2.0)[b]
		ADHD-vehicle 2.6 (0.8)	ADHD-vehicle 3.0 (0.9)	ADHD-vehicle 0.4 (0.1 to 0.7)[b]
	Level of good mood in general	ADHD-FA: 2.2 (1.1)	ADHD-FA: 3.7 (0.9)	ADHD-FA: 1.5 (1.2 to 1.8)[b]
		ADHD-vehicle 2.0 (0.9)	ADHD-vehicle 2.4 (1.0)	ADHD-vehicle 0.4 (0.1 to 0.7)[b]
	Level of ability to concentrate during the day	ADHD-FA: 2.0 (1.3)	ADHD-FA: 3.8 (0.8)	ADHD-FA: 1.8 (1.4 to 2.2)[b]
		ADHD-vehicle 1.9 (1.1)	ADHD-vehicle 2.4 (1.2)	ADHD-vehicle 0.5 (0.1 to 0.9)[b]
	Percentage of homework completed in general	ADHD-FA: 2.0 (1.3)	ADHD-FA: 3.3 (1.2)	ADHD-FA: 1.3 (0.9 to 1.7)[b]
		ADHD-vehicle 2.3 (1.5)	ADHD-vehicle 2.9 (1)	ADHD-vehicle 0.6 (0.2 to 1.0)[b]

ADHD-FA, attention deficit hyperactivity disorder – fatty acid; ADHD-vehicle, placebo comparator group of ADHD children.
a Estimated unadjusted difference between the intervention and control groups at follow-up.
b Change from pre to post intervention reported separately for the two groups.

TABLE 29 Adverse events in studies evaluating other non-pharmacological interventions

Study	Adverse events reported	Measures used
Francis and Demspter (2002)[156]	NR	
Gringras *et al.* (2014)[36]	No serious adverse events were reported Two-day skin rash, $n = 1$ (may have been related to the weighted blanket). The authors report that all other adverse events were unrelated illnesses such as colds, fever, chickenpox, broken bone in hand (number of children with other unrelated adverse events was not reported)	A 24-hour telephone number was available to parents for reporting adverse events Weekly parent reviews (face to face or telephone)
Guilleminault *et al.* (1993)[158]	NR. Heat caused by the artificial light was reported as the only complication to light therapy treatment	NR
Oriel *et al.* (2016)[159]	NR	
Piazza *et al.* (1997)[157]	NR	NR
Yehuda *et al.* (2011)[160]	Fatty acid supplement: transient stomach upset and diarrhoea, $n = 3$; dizziness, $n = 2$	NR
Yu and Hong (2012)[161]	NR	

NR, not reported.

Summary

Seven studies evaluated seven types of non-pharmacological interventions. These interventions were all different from each other. Therefore, we summarise the findings from each study in turn.

Weighted blankets were evaluated using a crossover RCT, with a non-weighted blanket as the control intervention.[36] The intervention was developed for children with ASD, and all children recruited to the study had this diagnosis. They were aged between 5 and 16 years. The trial was rated as having a high risk of bias. Outcomes were measured over each 2-week treatment period. Data on 27 children in each arm were analysed.

No benefits in terms of TST and SOL (derived from actigraphy) or a number of other sleep outcomes derived from parent-completed sleep diaries (number of wakings/night, proportion of nights with more than one waking and total time awake after sleep onset) were reported for children in the weighted blanket group compared with the non-weighted blanket group. Similarly, no benefits from a weighted blanket were reported in terms of child behaviour (ABC) or increased sensitivities to sensory stimulation (Sensory Behaviour Questionnaire). There were no serious adverse events.

Valerian, a herb prepared in tablet form, was evaluated with a placebo using a RCT[156] in a sample of five children with a range of NDs aged 7–14 years who had difficulties with sleep initiation and/or sleep maintenance. The rating of risk of bias was high.

At 2 weeks post intervention, there was no difference between arms in terms of TST, SOL and duration of time awake during the night. Parents' perceptions of sleep quality (measured using a non-standardised visual analogue scale) did not differ significantly between trial arms post intervention. No other quantitative data on child outcomes were reported. Adverse events were not reported.

Essential fatty acid supplement preparation in tablet form was evaluated using a controlled before-and-after study.[160] All children (active arm, $n = 40$; placebo, $n = 38$), aged 9–12 years, recruited to the study were diagnosed with ADHD and were described as 'sleep deprived'. The study was rated as having a high risk of bias.

At 10 weeks post intervention, scores on a non-standardised measure of sleep quality did not differ between active and placebo groups. A number of other parent-reported child outcomes were evaluated using non-standardised, parent-reported indicators (degree of fatigue in general during the day, level of good mood in general, level of ability to concentrate during the day and percentage of homework completed in general). No significant difference between active and placebo arms was found for these outcomes. Transient gastric problems were reported for some children in the active group.

An aquatic exercise programme for children with ASD (mean age 8.8 years) and 'sleep dysfunction' was evaluated using a before-and-after study design.[159] Eight children were recruited to the study. The study was rated as having a high risk of bias. The outcomes at 8 weeks post intervention were reviewed. Biweekly telephone interviews with parents were used to collect data, from which TST and SOL were calculated. Increased TST post intervention was reported (MD 83.3 minutes, 95% CI 17.55 to 149.09 minutes). No other changes to child sleep outcomes were reported. Adverse events were not reported.

Acupuncture and ear-point taping were also evaluated using a controlled before-and-after study design.[161] Thirty children (mean age 6.9 years) with sleep initiation or maintenance difficulties and/or an 'abnormal sleep state' were recruited to the study. All were described as having learning disabilities. It was not clear when post-intervention outcomes data were collected. The study was rated as having a high risk of bias. At post intervention, a significant improvement in scores on a parent-reported sleep outcome measure (CSHQ) was reported (MD –11.50, 95% CI –13.33 to –9.67).

A light therapy intervention combined with a programme of daytime schedules and activities was evaluated using a before-and-after study.[158] Fourteen children, aged 9 months to 4 years, were recruited to the study. They were described as having learning disabilities. The study was rated as having a high risk of bias. Outcomes were measured at 6 months post intervention. Five out of 14 children were reported to have 'responded to treatment' as measured by increases in TST (calculated from information collected by the study team during regular interviews with parents), which were reported to have improved post intervention. There were no serious adverse events.

Finally, the relative effectiveness of faded bedtime and response costs with bedtime scheduling was investigated using a parallel RCT.[157] The trial took place in an inpatient unit for children with very severe behaviour problems. The children ($n = 14$, 7 per arm) recruited to the trial had a range of NDs and sleep disturbances relating to sleep maintenance and 'sleep consolidation'. They were aged 4–14 years. The trial was rated as having a high risk of bias. Outcomes were measured pre intervention and 10 days post intervention. Only narrative reports of sleep outcomes (based on observational data) are provided.

Overall, there is no evidence of benefit for weighted blankets, although we note that this was a single study that was rated as having a high risk of bias. Issues regarding study design and study bias mean that no conclusions can be drawn regarding the impact of the following interventions: valerian, fatty acid supplements, light therapy with daytime activities programme, acupuncture and ear-point taping, and an aquatic exercise programme. Finally, we note that the last two studies described[157,158] were both reported almost 25 years ago, and no further studies replicating these interventions are reported. It is questionable whether or not they remain relevant or accepted approaches to managing sleep disturbance in children with NDs.

Issues of feasibility, acceptability and experiences of receiving and implementing a sleep management intervention

Sixteen of the interventions included in the clinical effectiveness review also investigated the feasibility, acceptability and/or parent/clinician views of sleep disturbance interventions.[36,49,106,107,122–130] Going forward, we use the term 'family experience' to refer to this topic area.

Fourteen of these interventions were parent-directed interventions,[21,49,107,123–129,149] the remaining two were evaluations of melatonin[106] and weighed blankets,[36] respectively. For 11 out of the 16 interventions, data on family experience are reported in papers alongside the presentation of data on outcomes. For one intervention,[149] these data are reported in a separate paper.[122] A further four interventions were collectively investigated with respect to the above issues and findings were reported separately to papers reporting intervention outcomes.[130]

No additional studies relevant to this topic were identified through searches. We would note that separate but relevant literature on parents' views and attitudes towards pharmacological and non-pharmacological interventions for sleep disturbance in children with NDs was identified (e.g. Goodday *et al.*,[180] Keenan *et al.*[181]). However, this body of work did not fall within the scope of this review, as these studies explored parental views of multiple sleep interventions.

A number of data collection methods were used, including questionnaires, structured interview and semistructured interviews (individual and group).

Ten studies[36,49,107,122–125,127–129] devised an intervention evaluation questionnaire to explore family experience. One of these studies also included semistructured interviews with a subsample.[124] A further study used a modified version of a previously published intervention evaluation questionnaire.[126] All collected data from parents, apart from one study that sought children's views about the weighted blanket they had been using.[36]

A further study used semistructured interviews to collect data from parents via individual interviews or groups.[130] Finally, one study reported data on reasons for study dropout. *Appendix 19* reports the methods used.[106]

Quality appraisal

The quality appraisal of 'family experience' studies used the tool created by Hawker *et al.*,[66] chosen because it was developed for use in reviews in which both qualitative and quantitative data are included and data in this review have been generated by studies using different paradigms. The tool was used in two ways. First, for studies ($n = 2$) in which a primary objective was to investigate family experience, and this is reported in a separate paper, a full quality appraisal was conducted (see *Appendix 20*). Second, for studies ($n = 10$) in which 'family experience' was a secondary study objective and was a minor element of evidence presented in any papers, the tool was used by the team to appraise, but not individually rate, the quality of the studies and evidence presented. One study[106] reported data that were a part of routine data collection within a trial (i.e. the reason for study dropout). We did not subject this specific element of the study to a separate quality appraisal.

For the 10 studies[36,49,107,123–129] for which 'family experience' was a secondary objective, the quality of studies and study reporting was very mixed. A number of issues were frequently noted that affected the quality of the 'family experience' objectives of these studies. First, family experience was sometimes not explicitly identified as a research objective. In addition, parent characteristics (e.g. gender, educational achievement, ethnicity and first language) were rarely reported. This is a significant omission, given that the majority of interventions represented in the 'family experience' data set were parent-directed interventions. Data collection tools were inadequately reported and/or study-specific tools were developed that, it would appear, had not undergone any piloting or been developed in consultation with parents. Descriptions of the analysis of family experience data were typically missing or very limited. Analyses were descriptive and opportunities to explore factors affecting experiences were not exploited. Based on the description of the data collection instruments, it appeared that, in some studies, not all findings were reported.

Findings

Findings on family experience can be found in *Appendix 19*.

Melatonin

One of the evaluations of melatonin reported that a child was withdrawn from the study owing to difficulties administering the medication to the child. No further data on family experience were identified with respect to the pharmacological interventions.[106]

Weighted blanket

Children taking part in the RCT evaluating weighted blankets were asked to describe their feelings about using the study blanket using a three-point scale ('really liked', 'just OK' or 'really disliked').[36] A greater proportion of children in the intervention group reported liking their blanket, but the differences in responses between the intervention and control groups were not significant.[36]

Parent-directed interventions

A number of themes, or topic areas, can be identified in the data reported:

- parents' experiences of accessing the intervention (e.g. time of day or competing demands on time)
- parents' experiences of implementing a sleep management strategy
- parents' views about the elements of the intervention that had an impact on the outcomes experienced
- parents' experiences as service users or of the process of receiving the intervention
- recommending the intervention to other parents.

It should be noted that some data that were identified as potentially relevant to these topic areas were ambiguous and open to different interpretations; for example, 'satisfaction' may refer to satisfaction with outcomes achieved or satisfaction with the way that an intervention is delivered. Data of this nature, although extracted and presented in *Appendix 19*, are not reported in the following narrative.

Parents' experiences of accessing the intervention

This topic was not widely or consistently explored. One study[124] reported that the majority of participants (five out of six) in their interview subsample believed that time pressures on parents and the time needed to make the initial commitment to embark on the intervention were barriers to parents accessing and completing such an intervention. In addition, one interviewee spontaneously mentioned that the time of day of the intervention was a 'least-liked' element of the intervention. A further study[126] reported that the time-consuming nature of the intervention was identified as something that was 'least liked' about the intervention they received. However, the authors do not clarify whether this refers to time attending sessions or the time required to implement a sleep management strategy. Johnson *et al.*[107] report data on very high levels of attendance at intervention sessions, which could be regarded as proxy evidence regarding the accessibility of the intervention.

Parents' experiences of implementing the sleep management strategy

Five studies report parents' views on the acceptability of the intervention, specifically the acceptability of the sleep management strategy that they were instructed to implement. Implementing new ways of managing sleep disturbance (e.g. not settling to sleep, night-time wakings) are likely to generate resistance and negative responses (e.g. crying) on the part of the child.

Bramble[122] reported that a minority (3/15) of their sample found this process 'rather tough' but were willing to continue. Two out of the twelve participants in Weiskop *et al.*[126] identified 'sticking to bedtime routine' as the least-liked element of the intervention. Austin *et al.*[123] reports that three out of five parents in their study found that implementing sleep management strategies was stressful. However, Moss *et al.*[124] reported that 21 out of 26 study participants described the implementation of the treatment plans as acceptable or very acceptable, and no improvements to the intervention were suggested. Both Austin *et al.*[123] and Weiskop *et al.*[126] also asked study participants if they would recommend the intervention to other parents and all said that they would.

Beresford *et al.*[130] describes parents' accounts of the demands that implementing, and sustaining, a sleep management strategy can place on them. A number of barriers to implementation were also identified. These included lack of consistency across caregivers, changes and disruptions in usual routines (e.g. owing to illness or holidays) and difficulties with the home environment, particularly if the child had to share with siblings.[130]

Adherence to implementing a sleep management strategy is an additional facet to this topic. Two studies provided relevant data.[107,125] First, Scibberas *et al.*[125] reported that 'Most caregivers reported that they could implement sleep management strategies "at least half of the time" '. No further details are provided. Second, Johnson *et al.*[107] collected clinicians' ratings of study participants' adherence to the intervention (including evidence of implementation of sleep management strategies) based on their observations during intervention sessions. In this study, an attention control arm was used; the intervention delivered to this arm was purely educational and did not require parents to implement behaviour management strategies. Over 90% adherence was reported for both study arms (active arm, 93%, 75–100%; control arm, 98%, 75–100%).

Parents' views about the elements of the intervention that had an impact on the outcomes experienced

Two approaches to exploring this topic were used. Some studies asked participants to rate the helpfulness, utility or relevance of a pre-set list of the different elements of the intervention they had received,[107,122,123,125,128] although this varied in terms of how fine-grained the elements were. Overall, there is consistent evidence that parents report the training delivered via the interventions as relevant and useful. It is not clear from the way that the data are reported whether or not parents vary in which elements they find particularly helpful. Differences in data collection instruments and ambiguities of language make further synthesis of the data impossible.

Other studies elicited parents' (spontaneous) views via interviews[124,127,130] or free-text responses in questionnaires.[126] Across these studies, the following were identified by parents as being important to supporting the achievement of positive sleep outcomes: receiving education on various aspects of sleep and sleep management,[124,130] training in specific strategies to manage problem behaviours (communication, behavioural or sensory),[124,127,130] that the intervention is tailored to the child,[124,130] provision of one-to-one instruction,[126] peer support via group delivery,[130] that attention is paid to developing parents' confidence regarding their parenting,[130] use of mechanisms for keeping track of progress (e.g. sleep diary)[130] and giving implementation support.[124,126,130]

Parents' experiences as service users or of the process of receiving the intervention

The mode of delivery is a core element of the service user experience. This was evaluated in one of the trials included in the review.[128] This study also captured data on family experience, including preference regarding mode of delivery. A smaller proportion of study participants in the individual delivery arm reported that they would have preferred the alternative mode of delivery than those in the group delivery arm (3/41 vs. 8/39).[128]

There were limited data on this topic area. A single participant (1/12) in the study by Weiskop *et al.*[126] found that the training sessions were too long. Reed *et al.*[129] reports that 10 out of 18 study participants believed that the duration of the intervention was sufficient.

Recommending the intervention to other parents

Five studies[123,126,128,129,144] specifically collected data on whether or not study participants would recommend the intervention to other parents. Three studies[123,126,129] reported that all parents receiving the intervention would recommend it to other parents. The other studies[128,144] reported that the great majority of study participants said that they would recommend the intervention. Malow *et al.*[128] compared the responses of parents receiving the intervention via individual face-to-face sessions with those receiving it via group

delivery. A greater proportion of study participants receiving the intervention face to face reported that they would recommend the intervention than those in the group delivery arm (38/41 vs. 32/39).[128]

Summary

Thirteen studies concerning family experience of sleep management interventions were identified, reporting 16 sleep management interventions in total.[36,49,106,107,122–130] All but one[106] of the studies concerned non-pharmacological interventions: 14 were parent-directed interventions[49,107,122–130] and one was a study of weighted blankets.[36] For these studies, data were predominantly quantitative (total $n = 225$) and typically collected using a questionnaire specifically designed for the study. One study[124] supplemented questionnaire data through semistructured interviews with a subsample ($n = 6$) of study participants. A further study[130] ($n = 35$), which investigated the family experience of four parent-directed sleep management interventions, used entirely qualitative methods. Study quality varied.

The pharmacological study[106] reported data relevant to family experience[106] concerning difficulties administering the medication. This was reported within the context of reporting on reasons for study withdrawal, rather than the study explicitly seeking to include an exploration of the family experience along with the evaluation of the medicine.[106]

Therefore, the most extensive data set concerns parents' experiences of receiving and implementing a sleep management intervention (11 studies[21,49,107,123–129,149]). The data were organised around five themes: parents' experiences of accessing the intervention, parents' experiences of implementing a sleep management strategy, parents' views about the elements of the intervention that had an impact on the outcomes experienced, parents' experiences as service users and recommending the intervention to others. Across all these themes, the data are very limited. There are a number of reasons for this. First, the pool of studies is very small. Second, many studies collected very few data. Third, studies typically used their own data collection instrument, thus hindering the pooling of data.

With these caveats, the following comments are made. There is very little evidence on parents' experiences of accessing sleep management interventions. One study reported that parents believed that the time requirements associated with receiving a sleep management intervention could be a barrier to parents accessing an intervention.[124] Another found that the time-consuming nature of the intervention was a feature of the intervention that was 'least liked' by some study participants.[126] However, across all studies, only one participant is reported as finding the duration of the intervention too long.[126]

Parents' experiences of implementing a sleep management strategy were explored by five studies.[107,122,123,126,130] A consistent theme across all – although it was not universally reported by study participants – is the challenges and demands placed on parents as they implement the strategy. Despite this, all of the studies that asked parents if they would recommend the intervention to others reported unanimous or high levels of recommendation. On a related point, a further study[144] reported exploring parents' judgements as to their adherence to a sleep management strategy and it suggests that consistent adherence to sleep management interventions should not be assumed. Another study identified a range of factors that may further interfere with implementing a sleep management strategy.[130]

Support with implementing a sleep management intervention was consistently identified by studies that offer data on elements of interventions that parents believe support positive outcomes. In terms of mode of delivery, both individual work and group delivery are identified as supporting positive outcomes: one offering the opportunity for a highly tailored intervention and the other offering peer support.

Chapter 4 Discussion

Introduction

Overall, the evidence on the management of sleep disturbance in children with NDs is very thin – in volume, scope and quality – particularly given that this group of conditions or diagnoses represents the majority of disabled children, and when the incidence and severity of sleep disturbance is greater than that for children with typical development. The conclusions that we have been able to draw regarding intervention effectiveness are limited to melatonin. Here, the conclusion is drawn that there is evidence of benefit; however, the clinical importance of the benefit is not certain. Therefore, there are no implications for health-care practice.

This lack of evidence is more compelling – as is the argument for strategic investment in this topic – given our understanding of the health, social and economic impacts of sleep deprivation. It is not surprising that in a national research prioritisation exercise for children with NDs, the management of sleep disturbance was ranked in the top 10 research priorities,[32] with both pharmacological and parent-directed interventions specified.

In this chapter, first, we present the strengths and limitations of the study and then public and patient involvement. Second, we discuss pharmacological interventions and principal findings with respect to this intervention approach. Third, we consider parent-directed interventions, providing an overview of these interventions before reporting principal findings and discussing the implications of these findings. Fourth, we move on to discuss other non-pharmacological interventions, presenting the principal findings with respect to this range of interventions. A final section discusses the issues and challenges for future research in this area.

Strengths and limitations of the study

We undertook thorough searches for eligible studies that included systematic searches of 16 databases, without language restrictions, and included sources for unpublished studies. We used standard methods, for example having two researchers undertake key study processes such as study selection, to reduce error and bias. The risk of bias in the included studies was assessed, although this assessment was often limited by poor reporting. We included small before-and-after studies that cannot provide a reliable estimate of the clinical effectiveness of an intervention because of the lack of a control group. These were included in order to help identify interventions that may be worth considering for evaluation in future RCTs. To militate against the limitations of some of the study designs included, we have clearly distinguished between randomised and non-randomised designs in the synthesis. We have also taken account of their limitations when drawing conclusions.

Given the number of interventions that were under consideration, a mixed-treatment comparison would have been the ideal statistical approach, permitting ranking of the benefits and harms of the different treatment options.[72] However, owing to the heterogeneity of the non-pharmacological studies, statistical pooling was not considered appropriate. As a result of the sparsity of combined and sequential interventions and poor reporting of any prior interventions received by participants in studies, a robust analysis of the impact that single, combined and sequential interventions had on clinical effectiveness was not possible. Similarly, any other planned subgroup analysis was possible only for the melatonin trials and this was limited by the small number of studies and the potential for confounding with other study characteristics.

It is not possible to blind the types of interventions and comparators used in the studies under consideration. In addition, owing to the nature of the outcomes measured, robust blinded outcome assessment is likely to be difficult. Although actigraphy-based child sleep outcomes are more objective than parent-reported

measures, we did not consider these to be true objective outcomes, with non-blinding being unlikely to have introduced bias. Therefore, all of the included non-pharmacological RCTs were rated as having a high risk of bias, even though blinding of treatment and comparator is not possible. For one study that compared a parent-directed tailored intervention with control,[138] this led to an overall rating of high risk of bias, despite all the other bias domains for that study being rating as having a low risk of bias. We acknowledge that applying the Cochrane criteria 'less strictly' would have led to this study having an overall rating of low risk of bias. This is an issue for all non-pharmacological studies in this area and we believe that it is unhelpful to have these studies rated as having a high risk of bias, as evidence from non-pharmacological interventions will always look weaker than that from pharmacological studies. Equally, we acknowledge that there is a risk of overestimating the clinical effectiveness of an intervention in which allocation is unblinded and outcomes have an element of subjectivity and may be influenced by lack of blinding. There is currently no established method of blinded outcome assessment in this field that we are aware of and further work in this area may be beneficial. There may also be value in further consideration by methodologists about how lack of blinding is graded in studies when blinding is not possible and the outcomes are subjective. Adoption of statistical approaches used in surgical studies, such as secondary statistical analyses taking into account participants' treatment allocation, may also have some value.[182]

Patient and public involvement

Three parents of children with ND (two mothers and one father) acted as project advisors. They were recruited from a permanent parent consultation group of the chief investigator's research unit. The children's diagnoses were autism and a rare, genetic condition.

The parents were invited to the project team meetings, which were held three times over the course of study and attended by the research team and all co-applicants. They were also occasionally consulted between meetings via e-mail.

Each parent attended at least one meeting. At the first meeting, an early item on the agenda was a presentation of an overview of systematic reviews as a research method. At the meetings, parents actively engaged in discussions. Their experiences also provided useful contextual information for the research team, some of whom had no prior experience of working in this topic area or with this particular group of children.

Pharmacological interventions

Research on the management of sleep disturbance affecting children with NDs is dominated by two intervention approaches: melatonin and parent-directed interventions. In terms of other pharmacological interventions, a small number of trials of other medicines were identified [acebutolol (Sectral®, Promius Pharma LLC, Princeton, NJ, USA), eszopiclone (Lunesta®, Sunovion Pharmaceuticals, Inc., Marlborough, MA, USA), gabapentin (Neurontin®, Pfizer Inc., New York City, NY, USA), ramelteon (Rozerem®, Takeda Pharmaceutical Company Ltd, Osaka, Japan) and zolpidem (Ambien®, Sanofi S.A., Paris, France)] but fell outside the eligibility criteria set for this review (these criteria were decided in consultation with clinicians). The review was, therefore, restricted to melatonin, clonidine and antihistamines. No studies of clonidine or antihistamines that fulfilled our study inclusion criteria were identified.

Principal findings: pharmacological interventions

There was evidence of benefit of melatonin compared with placebo, although the precise extent of the benefit, which children may benefit the most and the clinical importance of the benefit remain uncertain.

Discussion of principal findings: pharmacological interventions

This conclusion concurs with the issues raised by the team that conducted the only trial of (fast-release) melatonin rated as having a low risk of bias included in the review.[48] In it, the study sample comprised

children with developmental delay and some with additional diagnoses, including epilepsy, autism or a specific genetic or chromosomal disorder. The authors note that 'the sheer heterogeneity of the population studied has inevitably limited our ability to accurately estimate the impact of melatonin treatment for individual groups of patients with specific clinical (genetic), behavioural or developmental presentations'.[48]

A core inclusion criterion for the Appleton *et al.*[48] trial was that a parent-directed intervention – in the form of an advice booklet – had not successfully addressed the presenting sleep disturbance. For over half of the children recruited to the trial (56%), this intervention did not sufficiently address the sleep disturbance and they were randomised to the melatonin or placebo arms. This finding does lend support to the argument, and based on understandings of types and aetiologies of sleep disturbance in children with NDs, that pharmacological and non-pharmacological approaches may both be required. It can also be taken to suggest that for some types of sleep disturbance, recourse to melatonin should occur only once attempts to change the way that a family may be managing the sleep disturbance have been tried.

Parent-directed interventions

Behavioural insomnias (difficulties with bedtime settling and self-settling after night wakings) are defined as sleep patterns and behaviours developed through unhelpful sleep behaviours and sleep management practices.[183] Parent-directed interventions seek to provide parents with the knowledge and skills to change these patterns and behaviours.

This review adopted the term 'parent-directed interventions' to refer to sleep management interventions that involve training parents to respond to their child's sleep problems in different ways. The use of the term 'parent-directed' was deliberate. It emphasises that the direct recipient of the intervention is the parent, not the child. The child is merely the recipient of the outcomes the intervention achieves with respect to the parent. This has implications for the way in which such interventions should be understood and evaluated.

We categorised parent-directed interventions as comprehensive and non-comprehensive. Comprehensive interventions included training parents in evaluating the bedroom environment, paying attention to daytime and bedtime activities and routines (or sleep hygiene practices) and providing training in the use of particular behavioural strategies to manage specific problems related to settling and night waking. The majority of interventions were comprehensive, with two 'non-comprehensive' interventions also being included: one evaluating training on particular behaviour management strategies only[152] and the other providing sleep hygiene training for parents of children with ADHD.[155]

We made a further distinction with respect to these interventions: tailored versus non-tailored. Tailored interventions consisted of an assessment of the child's sleep and wider family context, which was then used to developed a personalised sleep management strategy. This approach necessarily meant that these interventions were delivered individually. They also all included implementation support, that is, contact between the practitioner and parent as the sleep management strategy in order to provide support or advice and/or suggest and supervise changes to the strategy.

Non-tailored interventions, in contrast, comprised the delivery of a standard 'training curriculum' in managing sleep disturbance in children with NDs. These were delivered via written material, in groups and/or individually. Some of these interventions had opportunities for parents to apply learning to their own child and/or implementation support.

This review identified some core intervention characteristics by which these interventions could be classified: comprehensive versus non-comprehensive; tailored versus non-tailored; mode(s) of delivery; number of practitioner–parent contacts; availability of implementation support; and condition-specific or generic ND.

Principal findings: parent-directed interventions

Studies of parent-directed interventions took two broad forms: (1) they were concerned with the clinical effectiveness of the intervention compared with no intervention or (2) they were testing different approaches to delivering the same intervention (e.g. varying mode of delivery or intensity of contact between the practitioner and the parent). There was variability between interventions in terms of intervention characteristics, such as mode of delivery, intensity, duration and availability of implementation support. Overall, the quality of the evidence was rated as having a high or unclear risk of bias in reporting and, therefore, the findings of these studies cannot be considered robust.

In terms of the tailored interventions, there was mixed evidence about the effects of these interventions on child (and parent) outcomes. There was limited evidence of benefit on more objective measures of sleep outcomes (actigraphy, verified or not with parent-completed sleep diaries). More evidence of benefit was found on parent-reported outcomes and a few parent outcomes. Conclusions are hampered by the limited number of RCTs, the multitude of outcome measures and the risk of studies being underpowered to detect an effect. Two small RCTs investigated specific intervention characteristics: mode of delivering implementation support[21] and a brief versus extended (with implementation support) version of an intervention.[125] The rating of a high risk of study bias and the trials being underpowered mean that no conclusions can be drawn.

Compared with the tailored interventions, the non-tailored interventions evaluated were even more diverse in terms of a number of intervention characteristics, including mode of delivery [i.e. written material, group delivery (single vs. multiple sessions) and one-to-one work], the extent to which they accommodated parents' individual learning and training needs and whether or not implementation support was available. Overall, the conclusions that can be drawn from this evidence are limited owing to the high risk of bias, the wide range in post-intervention follow-up time points and the risk of studies being underpowered to detect an effect. In addition, a large number of outcome measures were used, capturing a wide range of outcome domains.

There was mixed and very limited evidence regarding the impact that the provision of written information to parents had on managing their child's sleep disturbance. No conclusions can be drawn regarding the relative benefits of written versus face-to-face delivery of sleep management training to parents. There is some limited evidence of benefit for group-delivered interventions in terms of parent-reported sleep outcomes (but not reported for more objective measures of sleep). A single RCT[128] compared the individual delivery of an intervention via a single session with delivery of the same training via two group-delivered sessions. The rating of having a high risk of study bias and the trial being underpowered means that no conclusions can be drawn from the study findings. No RCTs were identified that evaluated a single-session workshop approach to delivering sleep management training to parents. It is useful to note that this mode of delivery is widely accessible to parents in the UK via statutory and non-statutory providers.

Two non-comprehensive parent-directed interventions are also reported by this study. The first is a trial (rated as having a high risk of bias) that evaluated an intervention in which the content was restricted to training on behavioural principles of managing problem behaviour compared with an attention control intervention.[152] The content of the second intervention was restricted to sleep hygiene principles and practices,[155] which was evaluated using a before-and-after study design.

Discussion of principal findings: parent-directed interventions

It is relevant to briefly contextualise these findings on parent-directed interventions within the wider evidence base on the clinical effectiveness of parent-directed interventions for managing sleep in children with typical development. Here, a recent review, which identified 15 trials[110,127,184–197] and 11 before-and-after studies,[149,198–208] concluded that there was 'moderate support' for such interventions for young children (≤ 5 years). A paucity of trials that included older children and adolescents limited any conclusions being drawn with respect to these older age groups.[184] The authors recommend that, for young children who have typical development and are healthy, parent-directed interventions should be implemented with

'no hesitation'. Furthermore, although noting the lack of evidence, the authors also recommend that they are used for older children and adolescents.

For children with NDs, however, additional factors may be at play in the development of behavioural insomnias, which may have implications for the content and scope of the training and advice given to parents, the duration of support parents may require to implement new sleep management approaches and strategies and the clinical effectiveness of a parent-directed intervention.[209] These include communication difficulties (both parental communication about bedtime and sleep and the child's ability to understand sleep cues or communicate their needs),[210,211] sensory sensitivities,[209] cognitive delay and arousal associated with daytime behaviour problems.[212] This has implications for the design of future evaluations.

Parent-directed interventions as complex interventions: implications

Parent-directed interventions to manage sleep disturbance in children with NDs can be regarded as 'complex intervention', that is, they comprise a number of different interconnected elements. Early guidance, issued by the Medical Research Council on developing and evaluating complex interventions,[213,214] has subsequently been developed and refined, including the need for intervention development and evaluation to be theory driven.[215] The theory underlying or informing an intervention will identify its 'active ingredients' and the factors that may moderate or mediate their therapeutic action. Although accepted as a fundamental feature of pharmacological studies, the notion of 'active ingredients' is as relevant and important to our understanding and evaluation of non-pharmacological interventions.[216,217] However, adoption of robust approaches to the evaluation of complex interventions, in which a theory of change has informed the study design and outcomes measured and the active ingredients are clearly defined and specified, remains patchy.[218]

Specifying a theory of change, and the active ingredients, to complex interventions such as parent-delivered sleep management interventions requires looking not only at content of the 'training' delivered to parents, per se, but also at the features of the individuals involved in delivery, at the context, or mode, in which the training is delivered and in the implementation.[216,219] It would be fair to say that the influence of this thinking and methodological debate has, to date, had relatively little impact on evaluations of parent-directed, sleep management interventions for children with NDs. Behavioural theories (e.g. extinction, positive reinforcement) are explicitly identified as underpinning all the sleep management strategies on which parents were trained in the comprehensive parent-directed interventions and one of the non-comprehensive interventions.[152] In addition, the influence of social learning theory[220] on choices about the techniques used to teach and train parents is apparent (e.g. the use of role-play, modelling, observation, group problem-solving). However, a theory of *the process* by which upskilling a parent in sleep management results in changes in a child's sleep, and the factors that have an impact on that process, is left (relatively) undiscussed and unexplored. This is evidenced by the fact that only a small minority of the studies measured changes in parents' knowledge of sleep management, perceived parenting confidence, collected data on parents' characteristics and/or sought to assess the extent to which parents accurately or consistently applied their training.

The evidence reviewed regarding the 'family experience' of parent-directed sleep management interventions offers some, limited, insights into the process by which parent-directed interventions affect changes in the duration or quality of a child's sleep and the factors that parents believe facilitate or hinder this process. However, the story appears complex. Thus, for example, although support with implementing new approaches to managing their child's sleep disturbance was consistently identified as a valued or helpful aspect of the intervention received by parents, views on mode of delivery revealed that there were different benefits in group versus individual delivery. Exposure to other parents' experiences and access to peer support were identified as supporting positive outcomes. In terms of individually delivered interventions, a key benefit reported by parents was the scope to personalise an intervention to the child and the family's specific needs and context. It is also important to note that parents are likely to vary in their abilities, capacities and/or willingness to access the different modes of delivery.[221]

Other non-pharmacological interventions

Seven other types of non-pharmacological intervention were included: valerian,[156] weighted blankets,[36] a sleep management intervention delivered in a specialist inpatient setting,[157] light therapy alongside a daytime activities programme,[158] an aquatic exercise programme,[159] acupuncture and ear-point taping [161] and fatty acid supplements.[160] The evaluations of two of these interventions – the sleep management intervention delivered in a specialist inpatient setting[157] and light therapy alongside a daytime activities programme[158] – were both carried out ≥ 20 years ago. We have included them in this review but their current relevance may be open to question.

Principal findings

There was no evidence that weighted blankets – an intervention that parents can purchase and use without professional supervision or receive via a prescription from statutory health services – were clinically effective. Issues of study design and ratings of high risk of study bias mean that no conclusions can be drawn regarding the impact of the following interventions: valerian, fatty acid supplements, light therapy with daytime activities programme, acupuncture and ear-point taping and the aquatic exercise programme.

Future research: overarching issues and challenges

A key aim of this review was to make recommendations regarding priorities for future research on this topic. In this section, we consider some overarching issues and challenges.

Outcomes and outcome measurement

A number of issues need to be addressed in future research to generate robust and meaningful evidence that, in future reviews, can be subject to meta-analysis.

First, a large number of different child sleep outcomes and measurement tools were used by the studies included in this review. Our synthesis of the studies was partly hampered by this diversity of outcome measures in relation to both child sleep outcomes and other outcomes for children and their parents. Only TST was measured in the majority of studies and assessed in a similar way across studies. We would note that when evidence of benefit of a sleep management intervention was reported – and actigraphy and parent-report measures were used – benefit was more frequently observed with respect to parent-reported measures. It is not clear whether this is because of issues of objectivity versus subjectivity in outcome measurement or that these different measurement approaches are capturing qualitatively different outcomes.

Adverse events were reported in the majority of melatonin trials. However, the type of adverse event reported, data collection methods used and reporting of adverse events varied. Standardisation of the reporting and data collection methods for adverse events in future trials is important to understand the safety of pharmacological interventions. This also has relevance for non-pharmacological studies, in which interventions may have unintended consequences.

There were two additional issues: (1) variability in the outcome domains of interest, for example carer outcomes were assessed in some studies and not others and (2) variability in the measure used to assess specific outcome domains. Previous research has identified sleep as a key health outcome for children and young people with ND, their parents[30] and health-care professionals.[222] However, based on searches of the Core Outcome Measures In Effectiveness database (www.comet-initiative.org; accessed 21 June 2017), no work has been undertaken to prioritise outcomes for children with NDs who experience sleep disturbances and their parents. A core outcome set would greatly assist the usefulness of research in this field, in which there are so many management options, allowing comparison between studies and also ensuring that the outcomes that are assessed are of relevance to children, their parents and also the health-care professionals involved in their care.

Second, further work with families to establish what constitutes a meaningful and worthwhile change in these outcome measures is important. Just one study[49] adopted this approach, with a 50% reduction in the composite sleep disturbance score identified by parents participating in the study as the minimum change for the intervention to be considered worthwhile.

Meaningful and sufficient change may also have guided the duration of implementation support in interventions in which the duration of this was reported as variable, or needs-led,[21,124,146] but the heuristic for ceasing implementation support was not explicitly reported. Furthermore, other studies reported an extended period of implementation support but it was not clear whether this duration was predetermined or simply what actually occurred when the intervention was delivered. An alternative, or additional, approach used by a small number of studies was the setting of parent-identified goals for their child's sleep, with progress towards these goals tracked over follow-up time points. However, none of these studies used Goal Attainment Scaling methodology,[223,224] which allows identification of, and tracking of progress towards, personalised intervention goals, with the creation of an aggregated score for each study participant, and allows robust comparisons between participants. This would appear to be a measurement approach worth considering in future research, alongside objective and other parent-reported outcome measures, as is the case in evaluations of complex interventions in other fields.[225]

Third, consistency in follow-up time points based on an evidence-informed theory of change is required. This is particularly an issue for non-pharmacological interventions in which the implementation of newly acquired knowledge and skills in managing a child's sleep may take time to have an effect. Thus, within the different types of parent-directed interventions, consensus is required as to the most clinically meaningful follow-up time points and whether or not evaluations should seek to also investigate maintenance of outcomes. This would also assist in comparisons between studies in future systematic reviews and meta-analyses.

Fourth, further work is required to identify other relevant child outcome measures *and* the time points at which such outcomes should be measured. Cognitive ability is an important example here. Little is understood about the benefits of (even small) cumulative gains in sleep in terms of arresting neuronal and cognitive loss.[226] Thus, the identification of appropriate follow-up time points and meaningful measures of cognition, an outcome that is challenging to accurately capture in children with severe learning disabilities, are essential. The selection of other child outcome measures also needs to be evidence based and informed by the intervention's theory of change for that outcome.

Fifth, for parent-directed interventions, the development and application of a theory of change is required in the selection of appropriate outcomes. This is because children's sleep outcomes will be mediated by the outcomes for parents achieved by the intervention, that is, the acquisition of new knowledge and understanding of sleep, and training in managing sleep disturbance. Furthermore, evidence from the wider literature on parenting interventions indicates that the extent to which these parent-centred outcomes are achieved are mediated/moderated by a number of factors, located in parent, practitioner and intervention characteristics. Only a minority of parent-directed interventions captured outcomes in this domain. Similarly, few studies report parent characteristics.

The design of evaluations

As we have noted earlier, evaluations need to be designed in such a way that the evidence generated addresses the key questions of what works, for whom and in what circumstances? Interestingly, a similar call for evidence has been made with respect to evaluation of similar interventions for children with typical development.[184] It would be informative to explore subgroup analyses within any future trials. In order to make any analyses more credible, it will be important to define these subgroups in advance, and the direction of effect, based on existing evidence.[227,228] Depending on the evaluation, subgroups may be in terms of sleep problem, child, parent or intervention characteristics.

In addition, for parent-directed interventions, RCTs need to incorporate measures of adherence and/or fidelity. These are loosely defined concepts. An evaluation of parent-directed interventions to manage autism

symptoms usefully defined it as both the accuracy and comprehensiveness of the intervention delivered to the parents by the practitioner and the consistency and competency of the parent as they implement new approaches to managing their child's sleep disturbance.[229] Our review of 'family experience' data identified that parents can find implementing a new sleep management strategy stressful and demanding. Only one study[125] attempted to assess this in any way, relying on parents' reports of the proportion of times that they implemented the new sleep management strategy. There are clearly significant challenges related to the feasibility and costs associated with measuring parent's adherence and fidelity to a sleep management strategy given the time of day when parents will be implementing a sleep management strategy and the home setting. Methodological research will be required to inform incorporating this into study designs.

Finally, the final follow-up time point for most studies allowed consideration of short-term outcomes only. Future studies need to consider what longer-term follow-ups should be incorporated into study designs. Evidence on longer-term outcomes is particularly pertinent for parent-directed interventions that require significant investment in parents', as well as practitioners', time, physical and emotional resources (and potentially entail significant, if short-term, disruption to the family). If available, such evidence would support shared decision-making regarding the management of a child's sleep disturbance.

Moving towards replication

At the moment, the evidence base predominantly comprises single evaluations of interventions. These are principally either investigations into the management of sleep disturbance by a particular clinic or are (typically) early evaluations of a newly developed intervention, prior to its implementation into routine practice. There are just two instances of some sort of replication study; however, in both,[123,124,128,129] there are differences between the studies in the way the intervention was delivered. A taxonomy by which interventions should be defined and adherence to Template for Intervention Description and Replication (TIDieR) guidance[164] in study reporting would support both replication and future evidence syntheses.

Chapter 5 Conclusions

Implications for health care

The poor quality of evidence and/or uncertainty regarding the clinical significance of findings mean that there are no implications for health-care practice.

Recommendations for research

Randomised controlled trials are required to assess a range of possible treatment options for sleep disturbance in children with ND. Future trials need to be informed by a greater understanding of the mechanisms by which non-pharmacological interventions may have an impact on a child's sleep and on outcomes and outcome measurement. Developing an understanding of these mechanisms will require mixed-methods research.

Future research needs to take account of the range of types, or aetiologies, of sleep disturbance, including behavioural, physiological (e.g. heightened arousal, anxiety or atypical melatonin profiles) and disorders of the circadian rhythm,[230] which will vary between NDs.

We would also highlight that reporting was poor across RCTs, before-and-after studies and in studies reporting parents' views and experiences of sleep management interventions. This was particularly the case for the non-pharmacological interventions. Future research should follow the appropriate reporting standards for the specific study design (www.equator-network.org; accessed 21 June 2017), including giving detailed description of the interventions using the TIDieR checklist.[164] Within the TIDieR checklist, it is important that sufficient detail is given on the specific curricula/training content of each intervention and the techniques used to teach and train parents.

Bearing all of these issues in mind, we recommend that the topic areas listed below are priorities for future research. Generating robust evidence in these topic areas will support clinicians (and families) to make evidence-informed decisions about the identification, prevention and management of sleep disturbance in children with NDs. It will also support evidence-informed decision-making with respect to commissioning of services. The suggested areas for future research are quite diverse. Although necessarily presented in an order, it would seem important that all key stakeholders contribute to any prioritisation processes.

- The development of a core outcome set would be beneficial. This should be developed in consultation with parents and carers, the children themselves, when possible, and health-care professionals and others involved in supporting parents and children, using a structured process such as that developed by the Core Outcome Measures In Effectiveness group.[231] Similarly, standardising the adverse events recorded, data collection methods used and level of detail reported is important for future evaluations of pharmacological and non-pharmacological interventions.
- If not already recently conducted, a review of existing tools, practices and strategies to identify sleep disturbance in children with NDs that are appropriate and feasible for use in routine practice is recommended.
- If available in a suitable preparation for the population under consideration, a trial comparing a slow-release formulation of melatonin with a fast-release formulation of melatonin is suggested.
- There are a range of possible other pharmacological interventions for this population, such as clonidine, but eligible studies were not identified. Prioritisation of evaluations of alternative pharmacological options to melatonin (e.g. clonidine) is recommended.

- No studies were identified that evaluated a combined or sequential use of melatonin (or other medicines) and parent-directed interventions. Studies of this nature are required because children may present with different types of sleep disturbance and/or sleep problems. Specifically, an apparent subclinical benefit of melatonin in managing sleep initiation difficulties may be beneficial in supporting the impact that a parent-directed intervention has. We recommend trials to investigate this.
- Parent-directed interventions range considerably in the intensity of practitioner input, from the simple provision of written material through to extended one-to-one contact and incorporation of implementation support. Studies are required that will support clinician and parent decision-making regarding the appropriate intervention. Evaluations of parent-directed interventions that allow for comparison of the relative impacts/clinical effectiveness of different features of the interventions (e.g. mode of delivery, group vs. individual and the nature of implementation support) are recommended, which should incorporate an investigation into issues of feasibility and acceptability. Prioritisation of interventions to address sleep initiation is suggested.
- None of the studies included in this review was presented as a preventative intervention. Rather, clinical cut-off points of sleep disturbance severity and/or parent reports of a sleep disturbance that had extended over some period of time were among study eligibility criteria. However, some types of sleep disturbance are preventable and/or amenable to early intervention. The brief, less-intense, parent-directed interventions included in this review (e.g. provision of information, single-session group intervention or face-to-face intervention) would, however, appear to align with a preventative or early intervention approach. Evaluating these types of intervention approaches in terms of the impact that they have on preventing the development of sleep disturbance, per se, or in preventing a newly emerging sleep disturbance increasing in severity is recommended.
- Studies that map current practices and provision and research into families' understanding of sleep disturbance and their experiences of seeking help are recommended. These would provide useful evidence to support the development of provision.

Finally, and across all topic areas, we would recommend that evaluations include a comprehensive and holistic economic evaluation, including costs to families.

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Contributions of authors

Bryony Beresford (Professor, Health and Care Services Research) was a joint co-applicant, contributed to all elements and, with Catriona McDaid, oversaw the delivery of the review and supervised junior members of the review team. She is joint lead author of this report.

Catriona McDaid (Senior Research Fellow, Systematic Reviews, Clinical Trials) was jointly responsible for writing the protocol and had shared responsibility for co-ordinating and leading the project, provided advice and input to all elements of the project and commented on drafts of the report.

Adwoa Parker (Research Fellow) contributed to study selection, data extraction, quality assessment and report writing.

Arabella Scantlebury (Research Fellow) contributed to study selection, data extraction, quality assessment and report writing.

Gemma Spiers (Research Fellow) worked on the project from February to September 2016. During that time, she was responsible for the day-to-day running of the project and led on screening, retrieval, data extraction and quality appraisal.

Caroline Fairhurst (Statistician) conducted the data analysis and contributed to the report writing.

Catherine Hewitt (Professor, Statistics) contributed to the protocol, provided methodological advice throughout the project and commented on drafts of the report.

Kath Wright (Research Fellow, Information Specialist) designed and undertook the literature searches and wrote related sections of the report.

Vicki Dawson (Founder and Chief Executive Officer, The Children's Sleep Charity) provided expert clinical advice throughout the project, contributed to screening and data extraction processes and to drafts of the report.

Heather Elphick (Consultant in Paediatric Respiratory and Sleep Medicine; Visiting Professor, Sheffield Hallam University) provided expert clinical advice throughout the project, contributed to screening and data extraction processes and to drafts of the report.

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Data-sharing statement

No primary data were produced. Most of the data extracted from the primary studies are available in the main body of the report and the appendices. Any further data can be obtained from the corresponding author.

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Appendix 1 Search strategies

Interface: Applied Social Sciences Index and Abstracts via ProQuest

Date range searched: no restriction.

Search date: 7 February 2016.

Records identified: 153.

Search strategy

((((SU.EXACT("Sleep disorders") OR SU.EXACT("Sleep problems")) OR SU.EXACT("Narcolepsy")) OR (("bed time*" NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (bedtime* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR ("bed time*" NEAR/3 (initial* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*)) OR ("bed time*" NEAR/3 (initial* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*))) OR ((night* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (night* NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*)) OR (nocturnal NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (nocturnal NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*))) OR ((sleep* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (sleep* NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*)) OR (sleep* NEAR/3 (initial* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*)) OR (sleepless* OR insomnia* OR parasomnia* OR "night terror*" OR nightterror* OR "night mare*" OR nightmare*) OR ("sleepwalk*" OR "nighthawk*" .) OR (sleepwalk* OR sleepwalk* OR "sleep walk*" OR somnambulism) OR (narcolepsy OR "nocturnal hyperkinesia"))) AND ((SU.EXACT("Children") OR SU.EXACT("adolescentce") OR SU.EXACT ("Infants")) OR (adolescent* OR baby OR babies OR child OR children OR boy OR boys OR girl OR girls OR infant* OR infancy* OR juvenile* OR paediatric OR pediatric OR preschooler* OR schoolboy* OR schoolgirl* OR schoolchild* OR teens OR teenager* OR toddler* OR youth OR youths OR "young people" OR "young person*"))) AND (((SU.EXACT("Developmentally disabled children") OR SU.EXACT("Developmentally delayed children")) OR (SU.EXACT("Developmental delays") OR SU.EXACT("Developmental disorders")) OR SU.EXACT ("Angelman syndrome") OR (SU.EXACT("Attention deficit disorder") OR SU.EXACT("Attention deficit hyperactivity disorder")) OR SU.EXACT("Conduct disorders") OR SU.EXACT("Complex partial seizure disorder" OR "Epilepsy" OR "Idiopathic childhood epilepsy" OR "Landau-Kleffner syndrome" OR "Panayiotopoulos syndrome" OR "Temporal lobe epilepsy") OR SU.EXACT("Cerebral palsy") OR SU.EXACT ("Down's syndrome") OR SU.EXACT("Fragile X syndrome") OR (SU.EXACT("Prader - Willi syndrome") OR SU.EXACT("Prader-Willi syndrome"))) OR (SU.EXACT("Rett syndrome") OR SU.EXACT("Smith-Magenis syndrome") OR (SU.EXACT("Williams-Beuren syndrome") OR SU.EXACT("Williams' syndrome")) OR (ADHD or "attention deficit" OR "angelman syndrome") OR (autism or autistic or asperges* OR "cerebral palsy") OR ("conduct disorder*" OR epilepsy or epileptic) OR ("Down* syndrome" OR "Fragile x syndrome") OR ("Prader Willi Syndrome" OR "Prader-Willi Syndrome") OR ("Rett syndrome" OR "Williams syndrome")) OR (developmental NEAR/2 (disability* OR delay*)) OR neurodisability* OR (neurodevelopment* NEAR/3 (delay* OR disability* OR disease* OR disorder* OR dysfunction)) OR (neuromotor* NEAR/3 (delay* OR disability* OR disease* OR disorder* OR dysfunction)) OR (neuropsychiatr* NEAR/3 (delay* OR disability* OR disease* OR disorder* OR dysfunction)) OR (neuropsychol* NEAR/3 (delay* OR disability* OR disease* OR disorder* OR dysfunction)))

Interface: Cumulative Index to Nursing & Allied Health via EBSCO*host*

Date range searched: no restriction.

Search date: 8 February 2016.

Records identified: 1157.

Search strategy

Search terms	Search options	Results
S37	S20 AND S36	1157
S36	S34 OR S35	66,730
S35	(developmental N2 (disabilit* or delay*)) OR neurodisabilit* OR (neurodevelopment* N3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) OR (neuromotor* N3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) OR (neuropsychiatric* N3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) OR (neuropsychol* N3 (delay* or disabilit* or disease* or disorder* or dysfunction*))	10,885
S34	(ADHD or (attention deficit)) OR (angelman syndrome) OR (autism or autistic or asperger*) OR (cerebral palsy) OR (conduct disorder*) OR (epilepsy or epileptic) OR (Down* syndrome) OR (Fragile x syndrome) OR (Prader Willi Syndrome) OR (Rett syndrome) OR (Smith-Magenis syndrome) OR (Williams syndrome)	58,518
S33	(MH "Williams Syndrome")	374
S32	(MH "Smith-Magenis Syndrome")	41
S31	(MH "Rett Syndrome")	374
S30	(MH "Prader-Willi Syndrome")	493
S29	(MH "Fragile X Syndrome")	673
S28	(MH "Down Syndrome")	5123
S27	(MH "Cerebral Palsy")	8165
S26	(MH "Epilepsy+")	11,511
S25	(MH "Child Behavior Disorders")	6418
S24	(MH "Attention Deficit Hyperactivity Disorder")	10,817
S23	(MH "Angelman Syndrome")	136
S22	(MH "Developmental Disabilities")	6552
S21	(MH "Child Development Disorders") OR (MH "Child Development Disorders, Pervasive")	3027
S20	S14 AND S19	9983
S19	S15 OR S16 OR S17 OR S18	793,040
S18	(adolescen* or baby or babies or child or children or boy or boys or girl or girls or infant* or infanc* or juvenile* or paediatric or pediatric or preschooler* or schoolboy* or schoolgirl* or schoolchild* or teens or teenager* or toddler* or youth or youths or young people or young person*)	793,040
S17	(MH "Adolescence")	350,551
S16	(MH "Infant")	118,046
S15	(MH "Child")	313,998
S14	S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7 OR S8 OR S9 OR S10 OR S11 OR S12 OR S13	41,776
S13	narcolepsy or nocturnal hyperkinesia	992

Search terms	Search options	Results
S12	((sleepless* or insomnia* or parasomnia* or night terror* or nightterror* or night mare* or nightmare*)) OR (sleep-wak* or night-wak*) OR (sleepwalk* or sleep-walk* or sleep walk* or somnambulism)	9309
S11	((sleep* N3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*)) OR ((sleep* N3 (dysfunction* or disorder or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR (sleep* N3 (initiat* or pattern* or routine* or practice* or maintain* or intervention* or schedule*))	17,544
S10	((nocturnal* N3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR ((nocturnal* N3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*))	366
S9	((night* N3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR ((night* N3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*))	996
S8	((bed time*) N3 (initiat* or pattern* or routine* or practice* or maintain* or intervention* or schedule*)) OR ((bedtime*) N3 (initiat* or pattern* or routine* or practice* or maintain* or intervention* or schedule*))	94
S7	((bed time*) N3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR ((bedtime*) N3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*))	66
S6	(MH "Dreams")	1111
S5	(MH "Parasomnias")	662
S4	(MH "Narcolepsy")	826
S3	(MH "Somnambulism")	151
S2	(MH "Sleep+")	16,169
S1	(MH "Sleep Disorders+")	26,313

Interface: The Cochrane Central Register of Controlled Trials via Wiley Online Library

Date range searched: no restriction.

Search date: 8 February 2016.

Records identified: 458.

Search strategy

#1 MeSH descriptor: [Sleep Wake Disorders] explode all trees

#2 MeSH descriptor: [Sleep] explode all trees

#3 MeSH descriptor: [Somnambulism] explode all trees

#4 MeSH descriptor: [Narcolepsy] explode all trees

#5 sleep* near/3 (initiat* or pattern* or routine* or practice* or maintain* or intervention* or schedule*):ti,ab,kw or sleepless* or insomnia* or parasomnia* or "night terror*" or nightterror* or "night mare*" or nightmare*:ti,ab,kw or "sleep-wak*" or "night-wak*":ti,ab,kw or "sleep-wak*" or "night-wak*" or sleepwalk* or "sleep-walk*" or "sleep walk*" or somnambulism:ti,ab,kw or narcolepsy or "nocturnal hyperkinesia":ti,ab,kw (Word variations have been searched)

#6 nocturnal near/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*):ti,ab,kw or nocturnal near/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*):ti,ab,kw or sleep* near/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*):ti,ab,kw or sleep* near/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*):ti,ab,kw (Word variations have been searched)

#7 night* near/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*):ti,ab,kw or night* near/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*):ti,ab,kw (Word variations have been searched)

#8 bed* near/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*):ti,ab,kw and bed* near/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*):ti,ab,kw (Word variations have been searched)

#9 #1 or #2 or #3 or #4 or #5 or #6 or #7 or #8

#10 MeSH descriptor: [Child] explode all trees

#11 MeSH descriptor: [Adolescent] explode all trees

#12 MeSH descriptor: [Infant] explode all trees

#13 adolescen* or baby or babies or child or children or boy or boys or girl or girls or infant* or infanc* or juvenile* or paediatric or pediatric or preschooler* or schoolboy* or schoolgirl* or schoolchild* or teens or teenager* or toddler* or youth or youths or "young people" or "young person*":ti,ab,kw (Word variations have been searched)

#14 #10 or #11 or #12 or #13

#15 #9 and #14

#16 MeSH descriptor: [Developmental Disabilities] explode all trees

#17 MeSH descriptor: [Child Development Disorders, Pervasive] explode all trees

#18 MeSH descriptor: [Angelman Syndrome] explode all trees

#19 MeSH descriptor: [Attention Deficit Disorder with Hyperactivity] explode all trees

#20 MeSH descriptor: [Attention Deficit and Disruptive Behavior Disorders] explode all trees

#21 MeSH descriptor: [Conduct Disorder] explode all trees

#22 MeSH descriptor: [Epilepsy] explode all trees

#23 MeSH descriptor: [Cerebral Palsy] explode all trees

#24 MeSH descriptor: [Down Syndrome] explode all trees

#25 MeSH descriptor: [Fragile X Syndrome] explode all trees

#26 MeSH descriptor: [Prader-Willi Syndrome] explode all trees

#27 MeSH descriptor: [Rett Syndrome] explode all trees

#28 MeSH descriptor: [Smith-Magenis Syndrome] explode all trees

#29 MeSH descriptor: [Williams Syndrome] explode all trees

#30 #16 or #17 or #18 or #19 or #20 or #21 or #22 or #23 or #24 or #25 or #26 or #27 or #28 or #29

#31 ADHD or "attention deficit":ti,ab,kw or "angelman syndrome":ti,ab,kw or autism or autistic or asperger*:ti,ab,kw or "cerebral palsy":ti,ab,kw or "conduct disorder*":ti,ab,kw (Word variations have been searched)

#32 epilepsy or epileptic:ti,ab,kw or "Down* syndrome":ti,ab,kw or "Fragile x syndrome":ti,ab,kw or "Prader Willi Syndrome":ti,ab,kw or "Rett syndrome":ti,ab,kw (Word variations have been searched)

#33 "Smith-Magenis syndrome":ti,ab,kw or "Williams syndrome":ti,ab,kw (Word variations have been searched)

#34 developmental near/2 (disabilit* or delay*):ti,ab,kw or neurodisabilit*:ti,ab,kw or neurodevelopment* near/3 (delay* or disabilit* or disease* or disorder* or dysfunction*):ti,ab,kw or neuromotor* near/3 (delay* or disabilit* or disease* or disorder* or dysfunction*):ti,ab,kw (Word variations have been searched)

#35 neuropsychiatric* near/3 (delay* or disabilit* or disease* or disorder* or dysfunction*):ti,ab,kw or neuropsychol* near/3 (delay* or disabilit* or disease* or disorder* or dysfunction*):ti,ab,kw (Word variations have been searched)

#36 #31 or #32 or #33 or #34 or #35

#37 #30 or #36

#38 #15 and #37

Interface: Cochrane Database of Systematic Reviews via Wiley Online Library

Date range searched: no restriction.

Search date: 8 February 2016.

Records identified: 8.

Search strategy: the Cochrane Database of Systematic Reviews strategy was the same as the CENTRAL strategy above.

Interface: Conference Proceedings Citation Index via Web of Science

Date range searched: no restriction.

Search date: 9 February 2016.

Records identified: 262.

Search strategy

#	Results	Search options
#13	262	#12 AND #9 *Indexes=CPCI-S Timespan=All years*
#12	28,755	#11 OR #10 *Indexes=CPCI-S Timespan=All years*
#11	2014	TOPIC: (developmental NEAR/2 (disabilit* or delay*)) *OR* TOPIC: (neurodisabilit*) *OR* TOPIC: (neurodevelopment* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) *OR* TOPIC: (neuromotor* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) *OR* TOPIC: (neuropsychiatric* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) *OR* TOPIC: (neuropsychol* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) *Indexes=CPCI-S Timespan=All years*
#10	27,174	TOPIC: (ADHD or "attention deficit") *OR* TOPIC: ("angelman syndrome") *OR* TOPIC: (autism or autistic or asperger*) *OR* TOPIC: ("cerebral palsy") *OR* TOPIC: ("conduct disorder*") *OR* TOPIC: (epilepsy or epilepti) *OR* TOPIC: ("Down* syndrome") *OR* TOPIC: ("Fragile x syndrome") *OR* TOPIC: ("Prader Willi Syndrome") *OR* TOPIC: ("Rett syndrome") *OR* TOPIC: ("Smith-Magenis syndrome") *OR* TOPIC: ("Williams syndrome") *Indexes=CPCI-S Timespan=All years*
#9	1224	#8 AND #7 *Indexes=CPCI-S Timespan=All years*
#8	146,852	TOPIC: (adolescen* or baby or babies or child or children or boy or boys or girl or girls or infant* or infanc* or juvenile* or paediatric or pediatric or preschooler* or schoolboy* or schoolgirl* or schoolchild* or teens or teenager* or toddler* or youth or youths or "young people" or "young person*") *Indexes=CPCI-S Timespan=All years*
#7	10,583	#6 OR #5 OR #4 OR #3 OR #2 OR #1 *Indexes=CPCI-S Timespan=All years*
#6	4957	TS=(sleep* NEAR/3 (initiat* or pattern* or routine* or practice* or maintain* or intervention* or schedule*)) OR TS=(sleepless* or insomnia* or parasomnia* or "night terror*" or nightterror* or "night mare*" or nightmare*) OR TS=(sleep-wak* or night-wak*) OR TS=(sleepwalk* or sleep-walk* or "sleep walk*" or somnambulism) OR TS=(narcolepsy or "nocturnal hyperkinesia") *Indexes=CPCI-S Timespan=All years*
#5	6374	TS=(nocturnal* NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(nocturnal* NEAR/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*)) OR TS=(sleep* NEAR/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*)) OR TS=(sleep* NEAR/3 (dysfunction* or disorder or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) *Indexes=CPCI-S Timespan=All years*
#4	393	TS=("bed time*" NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(bedtime* NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(night* NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(night* NEAR/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*)) *Indexes=CPCI-S Timespan=All years*
#3	641	TS=(narcolepsy) *Indexes=CPCI-S Timespan=All years*
#2	20	TS=(somnambulism) *Indexes=CPCI-S Timespan=All years*
#1	3818	TS=(sleep disorder*) *Indexes=CPCI-S Timespan=All years*

Interface: Database of Abstracts of Reviews of Effects via Wiley Online Library

Date range searched: no restriction.

Search date: 8 February 2016.

Records identified: 5.

Search strategy: the Database of Abstracts of Reviews of Effects strategy was the same as the CENTRAL strategy above.

Interface: EMBASE via Ovid

Date range searched: 1974 to 3 February 2016.

Search date: 4 February 2016.

Records identified: 10,288.

Search strategy

1. exp Sleep Disorder/ (162,095)
2. Sleep/ (81,239)
3. Sleep Walking/ (1490)
4. Narcolepsy/ (6862)
5. ((bed time or bedtime$) adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (318)
6. ((bed time$ or bedtime$) adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule$)).ti,ab. (282)
7. (night$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (2451)
8. (night$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (3053)
9. (nocturnal$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem $)).ti,ab. (1558)
10. (nocturnal$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (1279)
11. (sleep$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (22,635)
12. (sleep$ adj3 (dysfunction$ or disorder or disorders or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (50,363)
13. (sleep$ adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule $)).ti,ab. (13,975)
14. (sleepless$ or insomnia$ or parasomnia$ or night terror$ or nightterror$ or night mare$ or nightmare $).ti,ab. (29,033)
15. (sleep-wak$ or night-wak$).ti,ab. (11 to 232)
16. (sleepwalk$ or sleep-walk$ or sleep walk$ or somnambulism).ti,ab. (981)
17. (narcolepsy or nocturnal hyperkinesia).ti,ab. (5221)
18. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 (242,035)
19. exp Infant/ or exp Child/ or exp Adolescent/ (2,934,245)

20. (adolescen$ or baby or babies or child or children or boy or boys or girl or girls or infant$ or infanc$ or juvenile$ or paediatric or pediatric or preschooler$ or schoolboy$ or schoolgirl$ or schoolchild$ or teens or teenager$ or toddler$ or youth or youths or young people or young person$).ti,ab. (2,021,633)
21. 19 or 20 (3,492,441)
22. Childhood Disintegrative Disorder/ (128)
23. Developmental Disorder/ (28,670)
24. Asperger Syndrome/ (3660)
25. Attention Deficit Disorder/ or Attention Deficit Disorder with Hyperactivity/ or Attention Deficit Hyperactivity Disorder/ (43,642)
26. exp Autism/ (44,025)
27. Conduct Disorder/ (5234)
28. exp Epilepsy/ (194,464)
29. Cerebral Palsy/ (28,687)
30. Down Syndrome/ (29,198)
31. Fragile X Syndrome/ (6859)
32. Happy Puppet Syndrome/ (2139)
33. "Pervasive Developmental Disorder not otherwise specified"/ (790)
34. Prader-Willi Syndrome/ (4427)
35. Rett Syndrome/ (3903)
36. Smith Magenis Syndrome/ (480)
37. Williams Beuren Syndrome/ (2655)
38. (ADHD or attention deficit).ti,ab. (32,156)
39. angelman syndrome.ti,ab. (1370)
40. (autism or autistic or asperger$).ti,ab. (38,978)
41. cerebral palsy.ti,ab. (22,489)
42. conduct disorder$.ti,ab. (4762)
43. (epilepsy or epileptic).ti,ab. (137,265)
44. Down$ syndrome.ti,ab. (23,072)
45. Fragile x syndrome.ti,ab. (4278)
46. Prader Willi Syndrome.ti,ab. (3070)
47. Prader-Willi Syndrome.ti,ab. (3070)
48. Rett syndrome.ti,ab. (3072)
49. Smith-Magenis syndrome.ti,ab. (334)
50. Williams syndrome.ti,ab. (1688)
51. (developmental adj2 (disabilit$ or delay$)).ti,ab. (18,243)
52. neurodisabilit$.ti,ab. (271)
53. (neurodevelopment$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (8726)
54. (neuro-motor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (20)
55. (neuromotor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (428)
56. (neuropsychiatric$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (11,567)
57. (neuropsychol$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (2857)
58. 23 or 24 or 25 or 26 or 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51 or 52 or 53 or 54 or 55 or 56 or 57 (415,764)
59. 18 and 21 and 58 (10,326)
60. (animal/ or nonhuman/) not exp human/ (4,938,572)
61. 59 not 60 (10,288)

Interface: Health Management Information Consortium via Ovid

Date range searched: 1979 to November 2015.

Search date: 8 February 2016.

Records identified: 10.

Search strategy

1. ((bed time or bedtime$) adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (1)
2. ((bed time$ or bedtime$) adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule$)).ti,ab. (4)
3. (night$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (43)
4. (night$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (29)
5. (nocturnal$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem $)).ti,ab. (2)
6. (nocturnal$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (1)
7. (sleep$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (26)
8. (sleep$ adj3 (dysfunction$ or disorder or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (262)
9. (sleep$ adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule $)).ti,ab. (77)
10. (sleepless$ or insomnia$ or parasomnia$ or night terror$ or nightterror$ or night mare$ or nightmare $).ti,ab. (202)
11. (sleep-wak$ or night-wak$).ti,ab. (14)
12. (sleepwalk$ or sleep-walk$ or sleep walk$ or somnambulism).ti,ab. (1)
13. (narcolepsy or nocturnal hyperkinesia).ti,ab. (4)
14. (adolescen$ or baby or babies or child or children or boy or boys or girl or girls or infant$ or infanc$ or juvenile$ or paediatric or pediatric or preschooler$ or schoolboy$ or schoolgirl$ or schoolchild$ or teens or teenager$ or toddler$ or youth or youths or young people or young person$).ti,ab. (36 to 213)
15. (ADHD or attention deficit).ti,ab. (200)
16. angelman syndrome.ti,ab. (0)
17. (autism or autistic or asperger$).ti,ab. (546)
18. cerebral palsy.ti,ab. (144)
19. conduct disorder$.ti,ab. (85)
20. (epilepsy or epileptic).ti,ab. (372)
21. Down$ syndrome.ti,ab. (255)
22. Fragile x syndrome.ti,ab. (12)
23. Prader Willi Syndrome.ti,ab. (3)
24. Prader-Willi Syndrome.ti,ab. (3)
25. Rett syndrome.ti,ab. (5)
26. Smith-Magenis syndrome.ti,ab. (0)
27. Williams syndrome.ti,ab. (0)
28. (developmental adj2 (disabilit$ or delay$)).ti,ab. (100)
29. neurodisabilit$.ti,ab. (13)
30. (neurodevelopment$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (31)
31. (neuro-motor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (0)

32. (neuromotor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (0)
33. (neuropsychiatric$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (24)
34. (neuropsychol$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (7)
35. or/1-13 (559)
36. or/15-34 (1671)
37. 14 and 35 and 36 (10)

Interface: MEDLINE(R) In-Process & Other Non-Indexed Citations and Ovid MEDLINE(R) via Ovid

Date range searched: 1946 to present.

Search date: 4 February 2016.

Records identified: 4314.

Search strategy

1. exp Sleep Wake Disorders/ (67,333)
2. Sleep/ (41,262)
3. Somnambulism/ (561)
4. Narcolepsy/ (3034)
5. ((bed time or bedtime$) adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (175)
6. ((bed time$ or bedtime$) adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule$)).ti,ab. (146)
7. (night$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (1573)
8. (night$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (1850)
9. (nocturnal$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (1041)
10. (nocturnal$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (811)
11. (sleep$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (15,245)
12. (sleep$ adj3 (dysfunction$ or disorder or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (34,423)
13. (sleep$ adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule$)).ti,ab. (8810)
14. (sleepless$ or insomnia$ or parasomnia$ or night terror$ or nightterror$ or night mare$ or nightmare$).ti,ab. (17,408)
15. (sleep-wak$ or night-wak$).ti,ab. (7703)
16. (sleepwalk$ or sleep-walk$ or sleep walk$ or somnambulism).ti,ab. (665)
17. (narcolepsy or nocturnal hyperkinesia).ti,ab. (3478)
18. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 (125,134)
19. exp Infant/ or exp Child/ or Adolescent/ (3,027,976)
20. (adolescen$ or baby or babies or child or children or boy or boys or girl or girls or infant$ or infanc$ or juvenile$ or paediatric or pediatric or preschooler$ or schoolboy$ or schoolgirl$ or schoolchild$ or teens or teenager$ or toddler$ or youth or youths or young people or young person$).ti,ab. (1,603,721)
21. 19 or 20 (3,390,703)
22. exp Child Development Disorders, Pervasive/ (23,521)

23. Developmental Disabilities/ (16,203)
24. Angelman Syndrome/ (996)
25. exp "attention deficit and disruptive behavior disorders"/ or attention deficit disorder with hyperactivity/ or asperger syndrome/ or autistic disorder/ (42,213)
26. Conduct Disorder/ (2623)
27. exp Epilepsy/ (138,055)
28. Cerebral Palsy/ (16,902)
29. Down Syndrome/ (21,645)
30. Fragile X Syndrome/ (4267)
31. Prader-Willi Syndrome/ (2411)
32. Rett Syndrome/ (1997)
33. Smith-Magenis Syndrome/ (107)
34. Williams Syndrome/ (1371)
35. (ADHD or attention deficit).ti,ab. (22,948)
36. angelman syndrome.ti,ab. (1106)
37. (autism or autistic or asperger$).ti,ab. (29,066)
38. cerebral palsy.ti,ab. (16,339)
39. conduct disorder$.ti,ab. (3664)
40. (epilepsy or epileptic).ti,ab. (92,832)
41. Down$ syndrome.ti,ab. (18,536)
42. Fragile x syndrome.ti,ab. (3622)
43. Prader Willi Syndrome.ti,ab. (2414)
44. Prader-Willi Syndrome.ti,ab. (2414)
45. Rett syndrome.ti,ab. (2494)
46. Smith-Magenis syndrome.ti,ab. (285)
47. Williams syndrome.ti,ab. (1375)
48. (developmental adj2 (disabilit$ or delay$)).ti,ab. (13,041)
49. neurodisabilit$.ti,ab. (135)
50. (neurodevelopment$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (6427)
51. (neuro-motor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (11)
52. (neuromotor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (304)
53. (neuropsychiatric$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (8341)
54. (neuropsychol$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (2022)
55. 22 or 23 or 24 or 25 or 26 or 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51 or 52 or 53 or 54 (303,873)
56. 18 and 21 and 55 (4314)

Interface: PsycINFO via Ovid

Date range searched: 1806 to week 1 February 2016.

Search date: 4 February 2016.

Records identified: 1727.

Search strategy

1. exp Sleep Disorders/ (12,542)
2. Sleep/ (17,334)
3. Sleepwalking/ (384)
4. Narcolepsy/ (1251)
5. ((bed time or bedtime$) adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (184)

6. ((bed time$ or bedtime$) adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule$)).ti,ab. (154)
7. (night$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (972)
8. (night$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (1170)
9. (nocturnal$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (402)
10. (nocturnal$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (359)
11. (sleep$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (8609)
12. (sleep$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (17,897)
13. (sleep$ adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule$)).ti,ab. (5748)
14. (sleepless$ or insomnia$ or parasomnia$ or night terror$ or nightterror$ or night mare$ or nightmare$).ti,ab. (12,114)
15. (sleep-wak$ or night-wak$).ti,ab. (4320)
16. (sleepwalk$ or sleep-walk$ or sleep walk$ or somnambulism).ti,ab. (749)
17. (narcolepsy or nocturnal hyperkinesia).ti,ab. (1836)
18. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 (44,643)
19. (childhood birth 12 yrs or adolescence 13 17 yrs).ag. (657,693)
20. (adolescen$ or baby or babies or child or children or boy or boys or girl or girls or infant$ or infanc$ or juvenile$ or paediatric or pediatric or preschooler$ or schoolboy$ or schoolgirl$ or schoolchild$ or teens or teenager$ or toddler$ or youth or youths or young people or young person$).ti,ab. (760,813)
21. 19 or 20 (950,005)
22. "3250".cc. (35,406)
23. exp Pervasive Developmental Disorders/ (31,057)
24. exp Developmental Disabilities/ (12,448)
25. Neurodevelopmental Disorders/ (1374)
26. Attention Deficit Disorder/ (5012)
27. "Attention Deficit Disorder with Hyperactivity"/ (16,339)
28. exp Autism/ (23,157)
29. Aspergers Syndrome/ (2444)
30. Conduct Disorder/ (3750)
31. exp Epilepsy/ (22,293)
32. Cerebral Palsy/ (4098)
33. Down's Syndrome/ (5404)
34. Fragile X Syndrome/ (1368)
35. Prader Willi Syndrome/ (454)
36. Rett Syndrome/ (694)
37. Williams Syndrome/ (848)
38. (ADHD or attention deficit).ti,ab. (26,445)
39. angelman syndrome.ti,ab. (269)
40. (autism or autistic or asperger$).ti,ab. (37,207)
41. cerebral palsy.ti,ab. (5354)
42. conduct disorder$.ti,ab. (6181)
43. (epilepsy or epileptic).ti,ab. (29,858)
44. Down$ syndrome.ti,ab. (6294)
45. Fragile x syndrome.ti,ab. (1495)
46. Prader Willi Syndrome.ti,ab. (568)
47. Prader-Willi Syndrome.ti,ab. (568)

48. Rett syndrome.ti,ab. (887)
49. Smith-Magenis syndrome.ti,ab. (75)
50. Williams syndrome.ti,ab. (990)
51. (developmental adj2 (disabilit$ or delay$)).ti,ab. (10,810)
52. neurodisabilit$.ti,ab. (64)
53. (neurodevelopment$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (3603)
54. (neuro-motor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (5)
55. (neuromotor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (115)
56. (neuropsychiatric$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (5090)
57. (neuropsychol$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (2035)
58. 22 or 23 or 24 or 25 or 26 or 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51 or 52 or 53 or 54 or 55 or 56 or 57 (135,560)
59. 18 and 21 and 58 (1727)

Interface: Science Citation Index via Web of Science

Date range searched: no restriction.

Search date: 8 February 2016.

Records identified: 2831.

Search strategy

#	Results	Search options
#13	2831	#12 AND #9 *Indexes=SCI-EXPANDED Timespan=1900-2016*
#12	225,596	#11 OR #10 *Indexes=SCI-EXPANDED Timespan=1900-2016*
#11	31,246	TOPIC: (developmental NEAR/2 (disabilit* or delay*)) *OR* TOPIC: (neurodisabilit*) *OR* TOPIC: (neurodevelopment* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) *OR* TOPIC: (neuromotor* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) *OR* TOPIC: (neuropsychiatric* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) *OR* TOPIC: (neuropsychol* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) *Indexes=SCI-EXPANDED Timespan=1900-2016*
#10	203,758	TOPIC: (ADHD or "attention deficit") *OR* TOPIC: ("angelman syndrome") *OR* TOPIC: (autism or autistic or asperger*) *OR* TOPIC: ("cerebral palsy")*OR* TOPIC: ("conduct disorder*") *OR* TOPIC: (epilepsy or epilepti) *OR* TOPIC: ("Down* syndrome") *OR* TOPIC: ("Fragile x syndrome") *OR* TOPIC:("Prader Willi Syndrome") *OR* TOPIC: ("Rett syndrome") *OR* TOPIC: ("Smith-Magenis syndrome") *OR* TOPIC: ("Williams syndrome") *Indexes=SCI-EXPANDED Timespan=1900-2016*
#9	12,415	#8 AND #7 *Indexes=SCI-EXPANDED Timespan=1900-2016*
#8	1,497,016	TOPIC: (adolescen* or baby or babies or child or children or boy or boys or girl or girls or infant* or infanc* or juvenile* or paediatric or pediatric or preschooler* or schoolboy* or schoolgirl* or schoolchild* or teens or teenager* or toddler* or youth or youths or "young people" or "young person*") *Indexes=SCI-EXPANDED Timespan=1900-2016*
#7	76,127	#6 OR #5 OR #4 OR #3 OR #2 OR #1 *Indexes=SCI-EXPANDED Timespan=1900-2016*

#	Results	Search options
#6	36,633	TS=(sleep* NEAR/3 (initiat* or pattern* or routine* or practice* or maintain* or intervention* or schedule*)) OR TS=(sleepless* or insomnia* or parasomnia* or "night terror*" or nightterror* or "night mare*" or nightmare*) OR TS=(sleep-wak* or night-wak*) OR TS=(sleepwalk* or sleep-walk* or "sleep walk*" or somnambulism) OR TS=(narcolepsy or "nocturnal hyperkinesia") *Indexes=SCI-EXPANDED Timespan=1900-2016*
#5	47,890	TS=(nocturnal* NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(nocturnal* NEAR/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*)) OR TS=(sleep* NEAR/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*)) OR TS=(sleep* NEAR/3 (dysfunction* or disorder or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) *Indexes=SCI-EXPANDED Timespan=1900-2016*
#4	3852	TS=("bed time*" NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(bedtime* NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(night* NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(night* NEAR/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*)) *Indexes=SCI-EXPANDED Timespan=1900-2016*
#3	4917	TS=(narcolepsy) *Indexes=SCI-EXPANDED Timespan=1900-2016*
#2	297	TS=(somnambulism) *Indexes=SCI-EXPANDED Timespan=1900-2016*
#1	33,513	TS=(sleep disorder*) *Indexes=SCI-EXPANDED Timespan=All years*

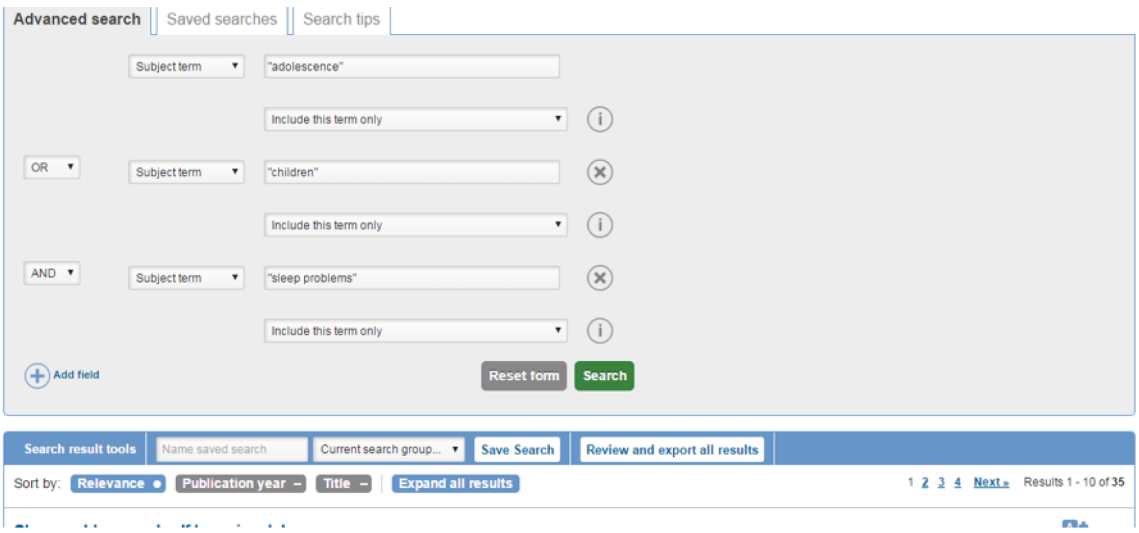
Interface: Social Care Online via www.scie-socialcareonline.org.uk

Date range searched: no restriction.

Search date: 4 February 2016.

Records identified: 35.

Search strategy



Interface: Social Policy & Practice via Ovid

Date range searched: no restriction.

Search date: 8 February 2016.

Records identified: 48.

Search strategy

1. ((bed time or bedtime$) adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (2)
2. ((bed time$ or bedtime$) adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule$)).ti,ab. (12)
3. (night$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (33)
4. (night$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (44)
5. (nocturnal$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem $)).ti,ab. (4)
6. (nocturnal$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (7)
7. (sleep$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (67)
8. (sleep$ adj3 (dysfunction$ or disorder or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (527)
9. (sleep$ adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule$)).ti,ab. (260)
10. (sleepless$ or insomnia$ or parasomnia$ or night terror$ or nightterror$ or night mare$ or nightmare$).ti,ab. (258)
11. (sleep-wak$ or night-wak$).ti,ab. (41)
12. (sleepwalk$ or sleep-walk$ or sleep walk$ or somnambulism).ti,ab. (13)
13. (narcolepsy or nocturnal hyperkinesia).ti,ab. (5)
14. (ADHD or attention deficit).ti,ab. (941)
15. angelman syndrome.ti,ab. (8)
16. (autism or autistic or asperger$).ti,ab. (2638)
17. cerebral palsy.ti,ab. (344)
18. conduct disorder$.ti,ab. (431)
19. (epilepsy or epileptic).ti,ab. (292)
20. Down$ syndrome.ti,ab. (509)
21. Fragile x syndrome.ti,ab. (49)
22. Prader Willi Syndrome.ti,ab. (32)
23. Prader-Willi Syndrome.ti,ab. (32)
24. Rett syndrome.ti,ab. (20)
25. Smith-Magenis syndrome.ti,ab. (3)
26. Williams syndrome.ti,ab. (12)
27. (developmental adj2 (disabilit$ or delay$)).ti,ab. (590)
28. neurodisabilit$.ti,ab. (19)
29. (neurodevelopment$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (64)
30. (neuro-motor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (0)
31. (neuromotor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (0)
32. (neuropsychiatric$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (32)
33. (neuropsychol$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (12)

34. or/1-13 (993)
35. or/14-33 (5507)
36. (adolescen$ or baby or babies or child or children or boy or boys or girl or girls or infant$ or infanc$ or juvenile$ or paediatric or pediatric or preschooler$ or schoolboy$ or schoolgirl$ or schoolchild$ or teens or teenager$ or toddler$ or youth or youths or young people or young person$).ti,ab. (139,979)
37. 34 and 35 and 36 (48)

Trial registers

In addition to the searches of the bibliographic databases, searches of the following trials registers were carried out: ClinicalTrials.gov, WHO International Clinical Trials Registry Platform and the UK Clinical Trials Gateway.

ClinicalTrials.gov via https://clinicaltrials.gov

This resource was searched on 9 February 2016 using a number of small focused search strategies. The results (103 records) were loaded into bibliographic software and, after deduplication, there were a total of 70 records. The strategies and numbers identified are given below:

- Sleep disorders & children & angelman (3)
- Sleep disorders & children & attention deficit/ADHD (13)
- Sleep disorders & children & autism (16)
- Sleep disorders & children & conduct disorder (46)
- Sleep disorders & children & epilepsy (10)
- Sleep disorders & children & down syndrome (5)
- Sleep disorders & children & cerebral palsy (1)
- Sleep disorders & children & fragile x syndrome (0)
- Sleep disorders & children & prader willi syndrome (2)
- Sleep disorders & children & Rett syndrome (1)
- Sleep disorders & children & Smith-Magenis syndrome (6)
- Sleep disorders & children & Williams syndrome (0)

The World Health Organization International Clinical Trials Registry Platform via http://apps.who.int/trialsearch/

This resource was searched on 9 February 2016 using the search terms ‘sleep AND children’ and 114 records for 108 trials were identified.

UK Clinical Trials Gateway via www.ukctg.nihr.ac.uk

This resource was searched on 9 February 2016 using the search phrase ‘sleep disorders and children’ and 16 trials were identified.

Appendix 2 Additional searches using terms not included in original search strategies

Interface: Applied Social Sciences Index and Abstracts via Proquest

Date range searched: no restriction.

Search date: 17 March 2016.

Records identified: 154.

Search strategy

Set number	Searched for	Databases	Results
S1	((((SU.EXACT("Sleep disorders") OR SU.EXACT("Sleep problems")) OR SU.EXACT ("Narcolepsy")) OR (("bed time*" NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (bedtime* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR ("bed time*" NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*)) OR ("bed time*" NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*))) OR ((night* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (night* NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*)) OR (nocturnal NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (nocturnal NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*))) OR ((sleep* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (sleep* NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*)) OR (sleep* NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*)) OR (sleepless* OR insomnia* OR parasomnia* OR "night terror*" OR nightterror* OR "night mare*" OR nightmare*) OR ("sleep-wak*" OR "night-wak*" .) OR (sleepwalk* OR sleep-walk* OR "sleep walk*" OR somnambulism) OR (narcolepsy OR "nocturnal hyperkinesia"))) AND ((SU.EXACT("Children") OR SU.EXACT("Adolescence") OR SU.EXACT("Infants")) OR (adolescen* OR baby OR babies OR child OR children OR boy OR boys OR girl OR girls OR infant* OR infanc* OR juvenile* OR paediatric OR pediatric OR preschooler* OR schoolboy* OR schoolgirl* OR schoolchild* OR teens OR teenager* OR toddler* OR youth OR youths OR "young people" OR "young person*"))) AND (((SU.EXACT("Developmentally disabled children") OR SU.EXACT("Developmentally delayed children")) OR (SU.EXACT("Developmental delays") OR SU.EXACT ("Developmental disorders")) OR SU.EXACT("Angelman syndrome") OR (SU.EXACT ("Attention deficit disorder") OR SU.EXACT("Attention deficit hyperactivity disorder")) OR SU.EXACT("Conduct disorders") OR SU.EXACT("Complex partial seizure disorder" OR "Epilepsy" OR "Idiopathic childhood epilepsy" OR "Landau-Kleffner syndrome" OR "Panayiotopoulos syndrome" OR "Temporal lobe epilepsy") OR SU.EXACT("Cerebral palsy") OR SU.EXACT("Down's syndrome") OR SU.EXACT("Fragile X syndrome") OR (SU.EXACT("Prader - Willi syndrome") OR SU.EXACT("Prader-Willi syndrome"))) OR (SU.EXACT("Rett syndrome") OR SU.EXACT("Smith-Magenis syndrome") OR (SU.EXACT ("Williams-Beuren syndrome") OR SU.EXACT("Williams' syndrome")) OR (ADHD or "attention deficit" OR "angelman syndrome") OR (autism or autistic or asperger* OR "cerebral palsy") OR ("conduct disorder*" OR epilepsy or epileptic) OR ("Down* syndrome" OR "Fragile x syndrome") OR ("Prader Willi Syndrome" OR "Prader-Willi Syndrome") OR ("Rett syndrome" OR "Williams syndrome")) OR (developmental NEAR/2 (disabilit* OR delay*)) OR neurodisabilit* OR (neurodevelopment* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuromotor* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuropsychiatr* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuropsychol* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)))	ASSIA	154

Set number	Searched for	Databases	Results
S2	(((SU.EXACT("Sleep disorders") OR SU.EXACT("Sleep problems")) OR SU.EXACT ("Narcolepsy")) OR (("bed time*" NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (bedtime* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR ("bed time*" NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*)) OR ("bed time*" NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*))) OR ((night* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (night* NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*)) OR (nocturnal NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (nocturnal NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*))) OR ((sleep* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (sleep* NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*)) OR (sleep* NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*)) OR (sleepless* OR insomnia* OR parasomnia* OR "night terror*" OR nightterror* OR "night mare*" OR nightmare*) OR ("sleep-wak*" OR "night-wak*" .) OR (sleepwalk* OR sleep-walk* OR "sleep walk*" OR somnambulism) OR (narcolepsy OR "nocturnal hyperkinesia")))	ASSIA	2877
S3	((SU.EXACT("Children") OR SU.EXACT("Adolescence") OR SU.EXACT("Infants")) OR (adolescen* OR baby OR babies OR child OR children OR boy OR boys OR girl OR girls OR infant* OR infanc* OR juvenile* OR paediatric OR pediatric OR preschooler* OR schoolboy* OR schoolgirl* OR schoolchild* OR teens OR teenager* OR toddler* OR youth OR youths OR "young people" OR "young person*"))	ASSIA	151,572
S4	(((SU.EXACT("Developmentally disabled children") OR SU.EXACT("Developmentally delayed children")) OR (SU.EXACT("Developmental delays") OR SU.EXACT ("Developmental disorders")) OR SU.EXACT("Angelman syndrome") OR (SU.EXACT ("Attention deficit disorder") OR SU.EXACT("Attention deficit hyperactivity disorder")) OR SU.EXACT("Conduct disorders") OR SU.EXACT("Complex partial seizure disorder" OR "Epilepsy" OR "Idiopathic childhood epilepsy" OR "Landau-Kleffner syndrome" OR "Panayiotopoulos syndrome" OR "Temporal lobe epilepsy") OR SU.EXACT("Cerebral palsy") OR SU.EXACT("Down's syndrome") OR SU.EXACT("Fragile X syndrome") OR (SU.EXACT("Prader - Willi syndrome") OR SU.EXACT("Prader-Willi syndrome"))) OR (SU.EXACT("Rett syndrome") OR SU.EXACT("Smith-Magenis syndrome") OR (SU.EXACT("Williams-Beuren syndrome") OR SU.EXACT("Williams' syndrome")) OR (ADHD or "attention deficit" OR "angelman syndrome") OR (autism or autistic or asperger* OR "cerebral palsy") OR ("conduct disorder*" OR epilepsy or epileptic) OR ("Down* syndrome" OR "Fragile x syndrome") OR ("Prader Willi Syndrome" OR "Prader-Willi Syndrome") OR ("Rett syndrome" OR "Williams syndrome")) OR (developmental NEAR/2 (disabilit* OR delay*)) OR neurodisabilit* OR (neurodevelopment* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuromotor* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuropsychiatr* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuropsychol* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)))	ASSIA	17,586
S6	(intellectual* NEAR/2 (disabilit* or disabled or deficit* or handicap* or retard*)) OR (mental* NEAR/2 (disabilit* or disabled or deficit* or handicap* or retard*)) OR (learning NEAR/2 (disability or disabled or difficult*))	ASSIA	11,198
S7	(((SU.EXACT("Sleep disorders") OR SU.EXACT("Sleep problems")) OR SU.EXACT ("Narcolepsy")) OR (("bed time*" NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (bedtime* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR ("bed time*" NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*)) OR ("bed time*" NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*))) OR ((night* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (night* NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*)) OR (nocturnal NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (nocturnal NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*))) OR	ASSIA These databases are searched for part of your query	154

Set number	Searched for	Databases	Results
	((sleep* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (sleep* NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*)) OR (sleep* NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*)) OR (sleepless* OR insomnia* OR parasomnia* OR "night terror*" OR nightterror* OR "night mare*" OR nightmare*) OR ("sleep-wak*" OR "night-wak*" .) OR (sleepwalk* OR sleep-walk* OR "sleep walk*" OR somnambulism) OR (narcolepsy OR "nocturnal hyperkinesia"))) AND ((SU.EXACT ("Children") OR SU.EXACT("Adolescence") OR SU.EXACT("Infants")) OR (adolescen* OR baby OR babies OR child OR children OR boy OR boys OR girl OR girls OR infant* OR infanc* OR juvenile* OR paediatric OR pediatric OR preschooler* OR schoolboy* OR schoolgirl* OR schoolchild* OR teens OR teenager* OR toddler* OR youth OR youths OR "young people" OR "young person*")) AND (((SU.EXACT ("Developmentally disabled children") OR SU.EXACT("Developmentally delayed children")) OR (SU.EXACT("Developmental delays") OR SU.EXACT("Developmental disorders")) OR SU.EXACT("Angelman syndrome") OR (SU.EXACT("Attention deficit disorder") OR SU.EXACT("Attention deficit hyperactivity disorder")) OR SU.EXACT ("Conduct disorders") OR SU.EXACT("Complex partial seizure disorder" OR "Epilepsy" OR "Idiopathic childhood epilepsy" OR "Landau-Kleffner syndrome" OR "Panayiotopoulos syndrome" OR "Temporal lobe epilepsy") OR SU.EXACT("Cerebral palsy") OR SU.EXACT("Down's syndrome") OR SU.EXACT("Fragile X syndrome") OR (SU.EXACT("Prader - Willi syndrome") OR SU.EXACT("Prader-Willi syndrome"))) OR (SU.EXACT("Rett syndrome") OR SU.EXACT("Smith-Magenis syndrome") OR (SU.EXACT("Williams-Beuren syndrome") OR SU.EXACT("Williams' syndrome")) OR (ADHD or "attention deficit" OR "angelman syndrome") OR (autism or autistic or asperger* OR "cerebral palsy") OR ("conduct disorder*" OR epilepsy or epileptic) OR ("Down* syndrome" OR "Fragile x syndrome") OR ("Prader Willi Syndrome" OR "Prader-Willi Syndrome") OR ("Rett syndrome" OR "Williams syndrome")) OR (developmental NEAR/2 (disabilit* OR delay*)) OR neurodisabilit* OR (neurodevelopment* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuromotor* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuropsychiatr* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuropsychol* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)))		
S8	(((SU.EXACT("Sleep disorders") OR SU.EXACT("Sleep problems")) OR SU.EXACT ("Narcolepsy")) OR (("bed time*" NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (bedtime* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR ("bed time*" NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*)) OR ("bed time*" NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*))) OR ((night* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (night* NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*)) OR (nocturnal NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (nocturnal NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*))) OR ((sleep* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (sleep* NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*)) OR (sleep* NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*)) OR (sleepless* OR insomnia* OR parasomnia* OR "night terror*" OR nightterror* OR "night mare*" OR nightmare*) OR ("sleep-wak*" OR "night-wak*" .) OR (sleepwalk* OR sleep-walk* OR "sleep walk*" OR somnambulism) OR (narcolepsy OR "nocturnal hyperkinesia"))) AND ((SU.EXACT ("Children") OR SU.EXACT("Adolescence") OR SU.EXACT("Infants")) OR (adolescen* OR baby OR babies OR child OR children OR boy OR boys OR girl OR girls OR infant* OR infanc* OR juvenile* OR paediatric OR pediatric OR preschooler* OR schoolboy* OR schoolgirl* OR schoolchild* OR teens OR teenager* OR toddler* OR youth OR youths OR "young people" OR "young person*")) AND ((((SU.EXACT ("Developmentally disabled children") OR SU.EXACT("Developmentally delayed children")) OR (SU.EXACT("Developmental delays") OR SU.EXACT("Developmental disorders")) OR SU.EXACT("Angelman syndrome") OR (SU.EXACT("Attention deficit disorder") OR SU.EXACT("Attention deficit hyperactivity disorder")) OR SU.EXACT ("Conduct disorders") OR SU.EXACT("Complex partial seizure disorder" OR	ASSIA These databases are searched for part of your query	174

Set number	Searched for	Databases	Results
	"Epilepsy" OR "Idiopathic childhood epilepsy" OR "Landau-Kleffner syndrome" OR "Panayiotopoulos syndrome" OR "Temporal lobe epilepsy") OR SU.EXACT("Cerebral palsy") OR SU.EXACT("Down's syndrome") OR SU.EXACT("Fragile X syndrome") OR (SU.EXACT("Prader - Willi syndrome") OR SU.EXACT("Prader-Willi syndrome"))) OR (SU.EXACT("Rett syndrome") OR SU.EXACT("Smith-Magenis syndrome") OR (SU.EXACT("Williams-Beuren syndrome") OR SU.EXACT("Williams' syndrome")) OR (ADHD or "attention deficit" OR "angelman syndrome") OR (autism or autistic or asperger* OR "cerebral palsy") OR ("conduct disorder*" OR epilepsy or epileptic) OR ("Down* syndrome" OR "Fragile x syndrome") OR ("Prader Willi Syndrome" OR "Prader-Willi Syndrome") OR ("Rett syndrome" OR "Williams syndrome")) OR (developmental NEAR/2 (disabilit* OR delay*)) OR neurodisabilit* OR (neurodevelopment* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuromotor* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuropsychiatr* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuropsychol* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction))) OR ((intellectual* NEAR/2 (disabilit* or disabled or deficit* or handicap* or retard*)) OR (mental* NEAR/2 (disabilit* or disabled or deficit* or handicap* or retard*)) OR (learning NEAR/2 (disability or disabled or difficult*))))		
S9	((((SU.EXACT("Sleep disorders") OR SU.EXACT("Sleep problems")) OR SU.EXACT ("Narcolepsy")) OR (("bed time*" NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (bedtime* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR ("bed time*" NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*)) OR ("bed time*" NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*))) OR ((night* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (night* NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*)) OR (nocturnal NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (nocturnal NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*))) OR ((sleep* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (sleep* NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*)) OR (sleep* NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*)) OR (sleepless* OR insomnia* OR parasomnia* OR "night terror*" OR nightterror* OR "night mare*" OR nightmare*) OR ("sleep-wak*" OR "night-wak*" .) OR (sleepwalk* OR sleep-walk* OR "sleep walk*" OR somnambulism) OR (narcolepsy OR "nocturnal hyperkinesia"))) AND ((SU.EXACT("Children") OR SU.EXACT ("Adolescence") OR SU.EXACT("Infants")) OR (adolescen* OR baby OR babies OR child OR children OR boy OR boys OR girl OR girls OR infant* OR infanc* OR juvenile* OR paediatric OR pediatric OR preschooler* OR schoolboy* OR schoolgirl* OR schoolchild* OR teens OR teenager* OR toddler* OR youth OR youths OR "young people" OR "young person*")) AND ((((SU.EXACT("Developmentally disabled children") OR SU.EXACT("Developmentally delayed children")) OR (SU.EXACT ("Developmental delays") OR SU.EXACT("Developmental disorders")) OR SU.EXACT ("Angelman syndrome") OR (SU.EXACT("Attention deficit disorder") OR SU.EXACT ("Attention deficit hyperactivity disorder")) OR SU.EXACT("Conduct disorders") OR SU.EXACT("Complex partial seizure disorder" OR "Epilepsy" OR "Idiopathic childhood epilepsy" OR "Landau-Kleffner syndrome" OR "Panayiotopoulos syndrome" OR "Temporal lobe epilepsy") OR SU.EXACT("Cerebral palsy") OR SU.EXACT("Down's syndrome") OR SU.EXACT("Fragile X syndrome") OR (SU.EXACT("Prader - Willi syndrome") OR SU.EXACT("Prader-Willi syndrome"))) OR (SU.EXACT("Rett syndrome") OR SU.EXACT("Smith-Magenis syndrome") OR (SU.EXACT("Williams-Beuren syndrome") OR SU.EXACT("Williams' syndrome")) OR (ADHD or "attention deficit" OR "angelman syndrome") OR (autism or autistic or asperger* OR "cerebral palsy") OR ("conduct disorder*" OR epilepsy or epileptic) OR ("Down* syndrome" OR "Fragile x syndrome") OR ("Prader Willi Syndrome" OR "Prader-Willi Syndrome") OR ("Rett syndrome" OR "Williams syndrome")) OR (developmental NEAR/2 (disabilit* OR delay*)) OR neurodisabilit* OR (neurodevelopment* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuromotor* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuropsychiatr* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuropsychol* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction))) OR ((intellectual*	ASSIA	20

Set number	Searched for	Databases	Results
	NEAR/2 (disabilit* or disabled or deficit* or handicap* or retard*)) OR (mental* NEAR/2 (disabilit* or disabled or deficit* or handicap* or retard*)) OR (learning NEAR/2 (disability or disabled or difficult*))))) NOT ((((SU.EXACT("Sleep disorders") OR SU.EXACT("Sleep problems")) OR SU.EXACT("Narcolepsy")) OR (("bed time*" NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (bedtime* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR ("bed time*" NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*)) OR ("bed time*" NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*))) OR ((night* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (night* NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*)) OR (nocturnal NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (nocturnal NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*))) OR ((sleep* NEAR/3 (dysfunction* OR disorder* OR difficult* OR disrupt* OR disturb* OR delay* OR problem*)) OR (sleep* NEAR/3 (settle* OR settling OR wake* OR awake OR wakeful* OR waking* OR awaking* OR awakening* OR wakening*)) OR (sleep* NEAR/3 (initiat* OR pattern* OR routine* OR practice* OR maintain* OR intervention* OR schedule*)) OR (sleepless* OR insomnia* OR parasomnia* OR "night terror*" OR nightterror* OR "night mare*" OR nightmare*) OR ("sleep-wak*" OR "night-wak*" .) OR (sleepwalk* OR sleep-walk* OR "sleep walk*" OR somnambulism) OR (narcolepsy OR "nocturnal hyperkinesia"))) AND ((SU.EXACT("Children") OR SU.EXACT("Adolescence") OR SU.EXACT("Infants")) OR (adolescen* OR baby OR babies OR child OR children OR boy OR boys OR girl OR girls OR infant* OR infanc* OR juvenile* OR paediatric OR pediatric OR preschooler* OR schoolboy* OR schoolgirl* OR schoolchild* OR teens OR teenager* OR toddler* OR youth OR youths OR "young people" OR "young person*")) AND (((SU.EXACT ("Developmentally disabled children") OR SU.EXACT("Developmentally delayed children")) OR (SU.EXACT("Developmental delays") OR SU.EXACT("Developmental disorders")) OR SU.EXACT("Angelman syndrome") OR (SU.EXACT("Attention deficit disorder") OR SU.EXACT("Attention deficit hyperactivity disorder")) OR SU.EXACT ("Conduct disorders") OR SU.EXACT("Complex partial seizure disorder" OR "Epilepsy" OR "Idiopathic childhood epilepsy" OR "Landau-Kleffner syndrome" OR "Panayiotopoulos syndrome" OR "Temporal lobe epilepsy") OR SU.EXACT("Cerebral palsy") OR SU.EXACT("Down's syndrome") OR SU.EXACT("Fragile X syndrome") OR (SU.EXACT("Prader - Willi syndrome") OR SU.EXACT("Prader-Willi syndrome"))) OR (SU.EXACT("Rett syndrome") OR SU.EXACT("Smith-Magenis syndrome") OR (SU.EXACT("Williams-Beuren syndrome") OR SU.EXACT("Williams' syndrome")) OR (ADHD or "attention deficit" OR "angelman syndrome") OR (autism or autistic or asperger* OR "cerebral palsy") OR ("conduct disorder*" OR epilepsy or epileptic) OR ("Down* syndrome" OR "Fragile x syndrome") OR ("Prader Willi Syndrome" OR "Prader-Willi Syndrome") OR ("Rett syndrome" OR "Williams syndrome")) OR (developmental NEAR/2 (disabilit* OR delay*)) OR neurodisabilit* OR (neurodevelopment* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuromotor* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuropsychiatr* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction)) OR (neuropsychol* NEAR/3 (delay* OR disabilit* OR disease* OR disorder* OR dysfunction))))		

Interface: The Cochrane Central Register of Controlled Trials via The Cochrane Library

Date range searched: no restriction.

Search date: 17 March 2016.

Records identified: 15.

Search strategy

#1 MeSH descriptor: [Sleep Wake Disorders] explode all trees

#2 MeSH descriptor: [Sleep] explode all trees

#3 MeSH descriptor: [Somnambulism] explode all trees

#4 MeSH descriptor: [Narcolepsy] explode all trees

#5 sleep* near/3 (initiat* or pattern* or routine* or practice* or maintain* or intervention* or schedule*):ti,ab,kw or sleepless* or insomnia* or parasomnia* or "night terror*" or nightterror* or "night mare*" or nightmare*:ti,ab,kw or "sleep-wak*" or "night-wak*":ti,ab,kw or "sleep-wak*" or "night-wak*" or sleepwalk* or "sleep-walk*" or "sleep walk*" or somnambulism:ti,ab,kw or narcolepsy or "nocturnal hyperkinesia":ti,ab,kw (Word variations have been searched)

#6 nocturnal near/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*):ti,ab,kw or nocturnal near/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*):ti,ab,kw or sleep* near/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*):ti,ab,kw or sleep* near/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*):ti,ab,kw (Word variations have been searched)

#7 night* near/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*):ti,ab,kw or night* near/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*):ti,ab,kw (Word variations have been searched)

#8 bed* near/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*):ti,ab,kw and bed* near/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*):ti,ab,kw (Word variations have been searched)

#9 #1 or #2 or #3 or #4 or #5 or #6 or #7 or #8

#10 MeSH descriptor: [Child] explode all trees

#11 MeSH descriptor: [Adolescent] explode all trees

#12 MeSH descriptor: [Infant] explode all trees

#13 adolescen* or baby or babies or child or children or boy or boys or girl or girls or infant* or infanc* or juvenile* or paediatric or pediatric or preschooler* or schoolboy* or schoolgirl* or schoolchild* or teens or teenager* or toddler* or youth or youths or "young people" or "young person*":ti,ab,kw (Word variations have been searched)

#14 #10 or #11 or #12 or #13

#15 #9 and #14

#16 MeSH descriptor: [Developmental Disabilities] explode all trees

#17 MeSH descriptor: [Child Development Disorders, Pervasive] explode all trees

#18 MeSH descriptor: [Angelman Syndrome] explode all trees

#19 MeSH descriptor: [Attention Deficit Disorder with Hyperactivity] explode all trees

#20 MeSH descriptor: [Attention Deficit and Disruptive Behavior Disorders] explode all trees

#21 MeSH descriptor: [Conduct Disorder] explode all trees

#22 MeSH descriptor: [Epilepsy] explode all trees

#23 MeSH descriptor: [Cerebral Palsy] explode all trees

#24 MeSH descriptor: [Down Syndrome] explode all trees

#25 MeSH descriptor: [Fragile X Syndrome] explode all trees

#26 MeSH descriptor: [Prader-Willi Syndrome] explode all trees

#27 MeSH descriptor: [Rett Syndrome] explode all trees

#28 MeSH descriptor: [Smith-Magenis Syndrome] explode all trees

#29 MeSH descriptor: [Williams Syndrome] explode all trees

#30 #16 or #17 or #18 or #19 or #20 or #21 or #22 or #23 or #24 or #25 or #26 or #27 or #28 or #29

#31 ADHD or "attention deficit":ti,ab,kw or "angelman syndrome":ti,ab,kw or autism or autistic or asperger*:ti,ab,kw or "cerebral palsy":ti,ab,kw or "conduct disorder*":ti,ab,kw (Word variations have been searched)

#32 epilepsy or epileptic:ti,ab,kw or "Down* syndrome":ti,ab,kw or "Fragile x syndrome":ti,ab,kw or "Prader Willi Syndrome":ti,ab,kw or "Rett syndrome":ti,ab,kw (Word variations have been searched)

#33 "Smith-Magenis syndrome":ti,ab,kw or "Williams syndrome":ti,ab,kw (Word variations have been searched)

#34 developmental near/2 (disabilit* or delay*):ti,ab,kw or neurodisabilit*:ti,ab,kw or neurodevelopment* near/3 (delay* or disabilit* or disease* or disorder* or dysfunction*):ti,ab,kw or neuromotor* near/3 (delay* or disabilit* or disease* or disorder* or dysfunction*):ti,ab,kw (Word variations have been searched)

#35 neuropsychiatric* near/3 (delay* or disabilit* or disease* or disorder* or dysfunction*):ti,ab,kw or neuropsychol* near/3 (delay* or disabilit* or disease* or disorder* or dysfunction*):ti,ab,kw (Word variations have been searched)

#36 #31 or #32 or #33 or #34 or #35

#37 #30 or #36

#38 #15 and #37

#39 MeSH descriptor: [Intellectual Disability] explode all trees

#40 intellectual* near/2 (disabilit* or disabled or deficit* or handicap* or retard*):ti,ab,kw (Word variations have been searched)

#41 (mental$ adj (disabilit$ or disabled or deficit$ or handicap$ or retard$))

#42 (learning adj (disability or disabled or difficult$))

#43 #39 or #40 or #41 or #42

#44 #37 or #43

#45 #9 and #14 and #44

#46 #45 not #38

Interface: Cumulative Index to Nursing & Allied Health via EBSCO*host*

Date range searched: no restriction.

Search date: 17 March 2016.

Records identified: 53.

Search strategy

Search identification number	Search terms	Results
S45	s44 NOT s42	53
S44	S14 AND S19 AND S43	1375
S43	S38 OR S39 OR S41	96,086
S42	S14 AND S19 AND S41	1322
S41	S21 OR S22 OR S23 OR S24 OR S25 OR S26 OR S27 OR S28 OR S29 OR S30 OR S31 OR S32 OR S33 OR S34 OR S35	74,324
S40	S38 OR S39	33,365
S39	(MH "Intellectual Disability+")	22,711
S38	(intellectual* N2 (disabilit* or disabled or deficit* or handicap* or retard*)) OR (mental* N2 (disabilit* or disabled or deficit* or handicap* or retard*)) OR (learning N2 (disabilit* or disabled or difficult*))	26,110
S37	S20 AND S36	Rerun
S36	S34 OR S35	Rerun
S35	(developmental N2 (disabilit* or delay*)) OR neurodisabilit* OR (neurodevelopment* N3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) OR (neuromotor* N3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) OR (neuropsychiatric* N3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) OR (neuropsychol* N3 (delay* or disabilit* or disease* or disorder* or dysfunction*))	Rerun
S34	(ADHD or (attention deficit)) OR (angelman syndrome) OR (autism or autistic or asperger*) OR (cerebral palsy) OR (conduct disorder*) OR (epilepsy or epileptic) OR (Down* syndrome) OR (Fragile x syndrome) OR (Prader Willi Syndrome) OR (Rett syndrome) OR (Smith-Magenis syndrome) OR (Williams syndrome)	Rerun
S33	(MH "Williams Syndrome")	Rerun
S32	(MH "Smith-Magenis Syndrome")	Rerun
S31	(MH "Rett Syndrome")	Rerun
S30	(MH "Prader-Willi Syndrome")	Rerun
S29	(MH "Fragile X Syndrome")	Rerun
S28	(MH "Down Syndrome")	Rerun
S27	(MH "Cerebral Palsy")	Rerun
S26	(MH "Epilepsy+")	Rerun

Search identification number	Search terms	Results
S25	(MH "Child Behavior Disorders")	Rerun
S24	(MH "Attention Deficit Hyperactivity Disorder")	Rerun
S23	(MH "Angelman Syndrome")	Rerun
S22	(MH "Developmental Disabilities")	Rerun
S21	(MH "Child Development Disorders") OR (MH "Child Development Disorders, Pervasive")	Rerun
S20	S14 AND S19	Rerun
S19	S15 OR S16 OR S17 OR S18	Rerun
S18	(adolescen* or baby or babies or child or children or boy or boys or girl or girls or infant* or infanc* or juvenile* or paediatric or pediatric or preschooler* or schoolboy* or schoolgirl* or schoolchild* or teens or teenager* or toddler* or youth or youths or young people or young person*)	Rerun
S17	(MH "Adolescence")	Rerun
S16	(MH "Infant")	Rerun
S15	(MH "Child")	Rerun
S14	S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7 OR S8 OR S9 OR S10 OR S11 OR S12 OR S13	Rerun
S13	narcolepsy or nocturnal hyperkinesia	Rerun
S12	((sleepless* or insomnia* or parasomnia* or night terror* or nightterror* or night mare* or nightmare*)) OR (sleep-wak* or night-wak*) OR (sleepwalk* or sleep-walk* or sleep walk* or somnambulism)	Rerun
S11	((sleep* N3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*)) OR ((sleep* N3 (dysfunction* or disorder or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR (sleep* N3 (initiat* or pattern* or routine* or practice* or maintain* or intervention* or schedule*))	Rerun
S10	((nocturnal* N3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR ((nocturnal* N3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*))	Rerun
S9	((night* N3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR ((night* N3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*))	Rerun
S8	((bed time*) N3 (initiat* or pattern* or routine* or practice* or maintain* or intervention* or schedule*)) OR ((bedtime*) N3 (initiat* or pattern* or routine* or practice* or maintain* or intervention* or schedule*))	Rerun
S7	((bed time*) N3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR ((bedtime*) N3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*))	Rerun
S6	(MH "Dreams")	Rerun
S5	(MH "Parasomnias")	Rerun
S4	(MH "Narcolepsy")	Rerun
S3	(MH "Somnambulism")	Rerun
S2	(MH "Sleep+")	Rerun
S1	(MH "Sleep Disorders+")	Rerun

Interface: Conference Proceedings Citation Index via Web of Science

Date range searched: no restriction.

Search date: 17 March 2016.

Records identified: 12.

Search strategy

#	Search options	Results
#17	#16 not #13	12
	Indexes=CPCI-S Timespan=All years	
#16	#15 AND #9	274
	Indexes=CPCI-S Timespan=All years	
#15	#14 OR #11 OR #10	31,261
	Indexes=CPCI-S Timespan=All years	
#14	TOPIC: (intellectual* NEAR/2 (disabilit* or disabled or deficit* or handicap* or retard*)) *OR* TOPIC: (mental* NEAR/2 (disabilit* or disabled or deficit* or handicap* or retard*)) *OR* TOPIC: (learning NEAR/2 (disability or disabled or difficult*))	3386
	Indexes=CPCI-S Timespan=All years	
#13	#12 AND #9	262
	Indexes=CPCI-S Timespan=All years	
#12	#11 OR #10	28,814
	Indexes=CPCI-S Timespan=All years	
#11	TOPIC: (developmental NEAR/2 (disabilit* or delay*)) *OR* TOPIC: (neurodisabilit*) *OR* TOPIC: (neurodevelopment* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) *OR* TOPIC: (neuromotor* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) *OR* TOPIC: (neuropsychiatric* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) *OR* TOPIC: (neuropsychol* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*))	2019
	Indexes=CPCI-S Timespan=All years	
#10	TOPIC: (ADHD or "attention deficit") *OR* TOPIC: ("angelman syndrome") *OR* TOPIC: (autism or autistic or asperger*) *OR* TOPIC: ("cerebral palsy") *OR* TOPIC: ("conduct disorder*") *OR* TOPIC: (epilepsy or epilepti) *OR* TOPIC: ("Down* syndrome") *OR* TOPIC: ("Fragile x syndrome") *OR* TOPIC: ("Prader Willi Syndrome") *OR* TOPIC: ("Rett syndrome") *OR* TOPIC: ("Smith-Magenis syndrome") *OR* TOPIC: ("Williams syndrome")	27,231
	Indexes=CPCI-S Timespan=All years	
#9	#8 AND #7	1227
	Indexes=CPCI-S Timespan=All years	
#8	TOPIC: (adolescen* or baby or babies or child or children or boy or boys or girl or girls or infant* or infanc* or juvenile* or paediatric or pediatric or preschooler* or schoolboy* or schoolgirl* or schoolchild* or teens or teenager* or toddler* or youth or youths or "young people" or "young person*")	148,172
	Indexes=CPCI-S Timespan=All years	
#7	#6 OR #5 OR #4 OR #3 OR #2 OR #1	10,610
	Indexes=CPCI-S Timespan=All years	

#	Search options	Results
#6	TS=(sleep* NEAR/3 (initiat* or pattern* or routine* or practice* or maintain* or intervention* or schedule*)) OR TS=(sleepless* or insomnia* or parasomnia* or "night terror*" or nightterror* or "night mare*" or nightmare*) OR TS=(sleep-wak* or night-wak*) OR TS=(sleepwalk* or sleep-walk* or "sleep walk*" or somnambulism) OR TS=(narcolepsy or "nocturnal hyperkinesia") *Indexes=CPCI-S Timespan=All years*	4969
#5	TS=(nocturnal* NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(nocturnal* NEAR/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*)) OR TS=(sleep* NEAR/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*)) OR TS=(sleep* NEAR/3 (dysfunction* or disorder or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) *Indexes=CPCI-S Timespan=All years*	6394
#4	TS=("bed time*" NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(bedtime* NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(night* NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(night* NEAR/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*)) *Indexes=CPCI-S Timespan=All years*	395
#3	TS=(narcolepsy) *Indexes=CPCI-S Timespan=All years*	641
#2	TS=(somnambulism) *Indexes=CPCI-S Timespan=All years*	20
#1	TS=(sleep disorder*) *Indexes=CPCI-S Timespan=All years*	3826

EMBASE via Ovid

Date range searched: 1974 to 16 March 2016.

Search date: 17 March 2016.

Records identified: 281.

Search strategy

1. exp Sleep Disorder/ (163,578)
2. Sleep/ (81,807)
3. Sleep Walking/ (1497)
4. Narcolepsy/ (6905)
5. ((bed time or bedtime$) adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (321)
6. ((bed time$ or bedtime$) adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule$)).ti,ab. (284)
7. (night$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (2464)
8. (night$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (3072)
9. (nocturnal$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem $)).ti,ab. (1566)

10. (nocturnal$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (1285)
11. (sleep$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (22,772)
12. (sleep$ adj3 (dysfunction$ or disorder or disorders or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (50,790)
13. (sleep$ adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule $)).ti,ab. (14,079)
14. (sleepless$ or insomnia$ or parasomnia$ or night terror$ or nightterror$ or night mare$ or nightmare $).ti,ab. (29,260)
15. (sleep-wak$ or night-wak$).ti,ab. (11,301)
16. (sleepwalk$ or sleep-walk$ or sleep walk$ or somnambulism).ti,ab. (987)
17. (narcolepsy or nocturnal hyperkinesia).ti,ab. (5254)
18. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 (244,169)
19. exp Infant/ or exp Child/ or exp Adolescent/ (2,954,891)
20. (adolescen$ or baby or babies or child or children or boy or boys or girl or girls or infant$ or infanc$ or juvenile$ or paediatric or pediatric or preschooler$ or schoolboy$ or schoolgirl$ or schoolchild$ or teens or teenager$ or toddler$ or youth or youths or young people or young person$).ti,ab. (2,038,598)
21. 19 or 20 (3,517,560)
22. Childhood Disintegrative Disorder/ (128)
23. Developmental Disorder/ (28,919)
24. Asperger Syndrome/ (3698)
25. Attention Deficit Disorder/ or Attention Deficit Disorder with Hyperactivity/ or Attention Deficit Hyperactivity Disorder/ (44,022)
26. exp Autism/ (44,576)
27. Conduct Disorder/ (5292)
28. exp Epilepsy/ (196,376)
29. Cerebral Palsy/ (29,042)
30. Down Syndrome/ (29,384)
31. Fragile X Syndrome/ (6913)
32. Happy Puppet Syndrome/ (2147)
33. "Pervasive Developmental Disorder not otherwise specified"/ (796)
34. Prader-Willi Syndrome/ (4459)
35. Rett Syndrome/ (3939)
36. Smith Magenis Syndrome/ (483)
37. Williams Beuren Syndrome/ (2672)
38. (ADHD or attention deficit).ti,ab. (32,439)
39. angelman syndrome.ti,ab. (1374)
40. (autism or autistic or asperger$).ti,ab. (39,494)
41. cerebral palsy.ti,ab. (22,786)
42. conduct disorder$.ti,ab. (4786)
43. (epilepsy or epileptic).ti,ab. (138,577)
44. Down$ syndrome.ti,ab. (23,196)
45. Fragile x syndrome.ti,ab. (4311)
46. Prader Willi Syndrome.ti,ab. (3090)
47. Prader-Willi Syndrome.ti,ab. (3090)
48. Rett syndrome.ti,ab. (3099)
49. Smith-Magenis syndrome.ti,ab. (335)
50. Williams syndrome.ti,ab. (1696)
51. (developmental adj2 (disabilit$ or delay$)).ti,ab. (18,431)
52. neurodisabilit$.ti,ab. (275)
53. (neurodevelopment$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (8887)

54. (neuro-motor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (21)
55. (neuromotor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (431)
56. (neuropsychiatric$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (11,690)
57. (neuropsychol$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (2874)
58. 23 or 24 or 25 or 26 or 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51 or 52 or 53 or 54 or 55 or 56 or 57 (419,846)
59. 18 and 21 and 58 (10,411)
60. (animal/ or nonhuman/) not exp human/ (4,980,395)
61. 59 not 60 (10,373)
62. intellectual impairment/ (14,389)
63. (intellectual$ adj (disabilit$ or disabled or deficit$ or handicap$ or retard$)).ti,ab. (13,554)
64. (mental$ adj (disabilit$ or disabled or deficit$ or handicap$ or retard$)).ti,ab. (44,139)
65. (learning adj (disability or disabled or difficult$)).ti,ab. (7378)
66. 62 or 63 or 64 or 65 (68,203)
67. 58 or 66 (462,906)
68. 18 and 21 and 67 (10,694)
69. (animal/ or nonhuman/) not exp human/ (4,980,395)
70. 68 not 69 (10,654)
71. 70 not 61 (281)

Interface: Health Management Information Centre via Ovid

Date range searched: 1979 to January 2016.

Search date: 17 March 2016.

Records identified: 4.

Search strategy

1. ((bed time or bedtime$) adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (1)
2. ((bed time$ or bedtime$) adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule$)).ti,ab. (4)
3. (night$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (43)
4. (night$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (29)
5. (nocturnal$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem $)).ti,ab. (2)
6. (nocturnal$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (1)
7. (sleep$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (26)
8. (sleep$ adj3 (dysfunction$ or disorder or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (262)
9. (sleep$ adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule $)).ti,ab. (77)
10. (sleepless$ or insomnia$ or parasomnia$ or night terror$ or nightterror$ or night mare$ or nightmare $).ti,ab. (202)
11. (sleep-wak$ or night-wak$).ti,ab. (14)

12. (sleepwalk$ or sleep-walk$ or sleep walk$ or somnambulism).ti,ab. (1)
13. (narcolepsy or nocturnal hyperkinesia).ti,ab. (4)
14. (adolescen$ or baby or babies or child or children or boy or boys or girl or girls or infant$ or infanc$ or juvenile$ or paediatric or pediatric or preschooler$ or schoolboy$ or schoolgirl$ or schoolchild$ or teens or teenager$ or toddler$ or youth or youths or young people or young person$).ti,ab. (36,281)
15. (ADHD or attention deficit).ti,ab. (200)
16. angelman syndrome.ti,ab. (0)
17. (autism or autistic or asperger$).ti,ab. (554)
18. cerebral palsy.ti,ab. (145)
19. conduct disorder$.ti,ab. (85)
20. (epilepsy or epileptic).ti,ab. (373)
21. Down$ syndrome.ti,ab. (255)
22. Fragile x syndrome.ti,ab. (12)
23. Prader Willi Syndrome.ti,ab. (3)
24. Prader-Willi Syndrome.ti,ab. (3)
25. Rett syndrome.ti,ab. (5)
26. Smith-Magenis syndrome.ti,ab. (0)
27. Williams syndrome.ti,ab. (0)
28. (developmental adj2 (disabilit$ or delay$)).ti,ab. (101)
29. neurodisabilit$.ti,ab. (13)
30. (neurodevelopment$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (31)
31. (neuro-motor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (0)
32. (neuromotor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (0)
33. (neuropsychiatric$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (24)
34. (neuropsychol$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (7)
35. or/1-13 (559)
36. or/15-34 (1680)
37. 14 and 35 and 36 (10)
38. (intellectual$ adj (disabilit$ or disabled or deficit$ or handicap$ or retard$)).ti,ab. (318)
39. (mental$ adj (disabilit$ or disabled or deficit$ or handicap$ or retard$)).ti,ab. (2397)
40. (learning adj (disability or disabled or difficult$)).ti,ab. (2394)
41. 38 or 39 or 40 (4883)
42. 36 or 41 (6387)
43. 14 and 35 and 42 (14)
44. 43 not 37 (4)

Interface: MEDLINE via Ovid Ovid MEDLINE(R) In-Process & Other Non-Indexed Citations and Ovid MEDLINE(R)

Date range searched: 1946 to present.

Search date: 17 March 2016.

Records identified: 220.

Search strategy

1. exp Sleep Wake Disorders/ (68,198)
2. Sleep/ (41,721)
3. Somnambulism/ (563)
4. Narcolepsy/ (3053)

5. ((bed time or bedtime$) adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (182)
6. ((bed time$ or bedtime$) adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule$)).ti,ab. (149)
7. (night$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (1593)
8. (night$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (1872)
9. (nocturnal$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (1051)
10. (nocturnal$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (819)
11. (sleep$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (15,445)
12. (sleep$ adj3 (dysfunction$ or disorder or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (35,005)
13. (sleep$ adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule$)).ti,ab. (8934)
14. (sleepless$ or insomnia$ or parasomnia$ or night terror$ or nightterror$ or night mare$ or nightmare$).ti,ab. (17,638)
15. (sleep-wak$ or night-wak$).ti,ab. (7799)
16. (sleepwalk$ or sleep-walk$ or sleep walk$ or somnambulism).ti,ab. (668)
17. (narcolepsy or nocturnal hyperkinesia).ti,ab. (3509)
18. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 (126,691)
19. exp Infant/ or exp Child/ or Adolescent/ (3,049,474)
20. (adolescen$ or baby or babies or child or children or boy or boys or girl or girls or infant$ or infanc$ or juvenile$ or paediatric or pediatric or preschooler$ or schoolboy$ or schoolgirl$ or schoolchild$ or teens or teenager$ or toddler$ or youth or youths or young people or young person$).ti,ab. (1,619,190)
21. 19 or 20 (3,417,108)
22. exp Child Development Disorders, Pervasive/ (23,981)
23. Developmental Disabilities/ (16,408)
24. Angelman Syndrome/ (1005)
25. exp "attention deficit and disruptive behavior disorders"/ or attention deficit disorder with hyperactivity/ or asperger syndrome/ or autistic disorder/ (42,816)
26. Conduct Disorder/ (2671)
27. exp Epilepsy/ (138,933)
28. Cerebral Palsy/ (16,993)
29. Down Syndrome/ (21,762)
30. Fragile X Syndrome/ (4312)
31. Prader-Willi Syndrome/ (2429)
32. Rett Syndrome/ (2027)
33. Smith-Magenis Syndrome/ (115)
34. Williams Syndrome/ (1394)
35. (ADHD or attention deficit).ti,ab. (23,416)
36. angelman syndrome.ti,ab. (1121)
37. (autism or autistic or asperger$).ti,ab. (29,748)
38. cerebral palsy.ti,ab. (16,486)
39. conduct disorder$.ti,ab. (3715)
40. (epilepsy or epileptic).ti,ab. (93,799)
41. Down$ syndrome.ti,ab. (18,660)
42. Fragile x syndrome.ti,ab. (3664)
43. Prader Willi Syndrome.ti,ab. (2435)
44. Prader-Willi Syndrome.ti,ab. (2435)

45. Rett syndrome.ti,ab. (2537)
46. Smith-Magenis syndrome.ti,ab. (286)
47. Williams syndrome.ti,ab. (1389)
48. (developmental adj2 (disabilit$ or delay$)).ti,ab. (13,253)
49. neurodisabilit$.ti,ab. (143)
50. (neurodevelopment$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (6630)
51. (neuro-motor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (11)
52. (neuromotor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (304)
53. (neuropsychiatric$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (8556)
54. (neuropsychol$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (2050)
55. 22 or 23 or 24 or 25 or 26 or 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51 or 52 or 53 or 54 (307,168)
56. 18 and 21 and 55 (4385)
57. Intellectual Disability/ (49,399)
58. (intellectual$ adj (disabilit$ or disabled or deficit$ or handicap$ or retard$)).ti,ab. (9558)
59. (mental$ adj (disabilit$ or disabled or deficit$ or handicap$ or retard$)).ti,ab. (34,226)
60. (learning adj (disability or disabled or difficult$)).ti,ab. (5309)
61. or/57-60 (74,629)
62. 55 or 61 (359,076)
63. 18 and 21 and 62 (4605)
64. 63 not 56 (220)

Interface: PsycINFO via Ovid

Date range searched: 1806 to week 2 March 2016.

Search date: 17 March 2016.

Records identified: 135.

Search strategy

1. exp Sleep Disorders/ (12,601)
2. Sleep/ (17,435)
3. Sleepwalking/ (384)
4. Narcolepsy/ (1260)
5. ((bed time or bedtime$) adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (186)
6. ((bed time$ or bedtime$) adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule$)).ti,ab. (155)
7. (night$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (978)
8. (night$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (1173)
9. (nocturnal$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (402)
10. (nocturnal$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (359)
11. (sleep$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (8660)

12. (sleep$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (18,021)
13. (sleep$ adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule $)).ti,ab. (5781)
14. (sleepless$ or insomnia$ or parasomnia$ or night terror$ or nightterror$ or night mare$ or nightmare$).ti,ab. (12,168)
15. (sleep-wak$ or night-wak$).ti,ab. (4347)
16. (sleepwalk$ or sleep-walk$ or sleep walk$ or somnambulism).ti,ab. (751)
17. (narcolepsy or nocturnal hyperkinesia).ti,ab. (1850)
18. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 (44,891)
19. (childhood birth 12 yrs or adolescence 13 17 yrs).ag. (660,440)
20. (adolescen$ or baby or babies or child or children or boy or boys or girl or girls or infant$ or infanc$ or juvenile$ or paediatric or pediatric or preschooler$ or schoolboy$ or schoolgirl$ or schoolchild$ or teens or teenager$ or toddler$ or youth or youths or young people or young person$).ti,ab. (764,560)
21. 19 or 20 (954,409)
22. "3250".cc. (35,699)
23. exp Pervasive Developmental Disorders/ (701)
24. exp Developmental Disabilities/ (12,490)
25. Neurodevelopmental Disorders/ (1401)
26. Attention Deficit Disorder/ (5017)
27. "Attention Deficit Disorder with Hyperactivity"/ (16,484)
28. exp Autism/ (0)
29. Aspergers Syndrome/ (0)
30. Conduct Disorder/ (3766)
31. exp Epilepsy/ (22,533)
32. Cerebral Palsy/ (4159)
33. Down's Syndrome/ (5416)
34. Fragile X Syndrome/ (1379)
35. Prader Willi Syndrome/ (454)
36. Rett Syndrome/ (701)
37. Williams Syndrome/ (853)
38. (ADHD or attention deficit).ti,ab. (26,638)
39. angelman syndrome.ti,ab. (269)
40. (autism or autistic or asperger$).ti,ab. (37,492)
41. cerebral palsy.ti,ab. (5404)
42. conduct disorder$.ti,ab. (6208)
43. (epilepsy or epileptic).ti,ab. (30,119)
44. Down$ syndrome.ti,ab. (6310)
45. Fragile x syndrome.ti,ab. (1506)
46. Prader Willi Syndrome.ti,ab. (572)
47. Prader-Willi Syndrome.ti,ab. (572)
48. Rett syndrome.ti,ab. (891)
49. Smith-Magenis syndrome.ti,ab. (75)
50. Williams syndrome.ti,ab. (998)
51. (developmental adj2 (disabilit$ or delay$)).ti,ab. (10,871)
52. neurodisabilit$.ti,ab. (66)
53. (neurodevelopment$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (3656)
54. (neuro-motor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (5)
55. (neuromotor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (115)
56. (neuropsychiatric$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (5141)
57. (neuropsychol$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (2044)

58. 22 or 23 or 24 or 25 or 26 or 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51 or 52 or 53 or 54 or 55 or 56 or 57 (136,113)
59. 18 and 21 and 58 (1739)
60. exp intellectual development disorder/ (40,609)
61. (intellectual$ adj (disabilit$ or disabled or deficit$ or handicap$ or retard$)).ti,ab. (10,964)
62. (mental$ adj (disabilit$ or disabled or deficit$ or handicap$ or retard$)).ti,ab. (28,202)
63. (learning adj (disability or disabled or difficult$)).ti,ab. (14,016)
64. 60 or 61 or 62 or 63 (65,071)
65. 58 or 64 (183,477)
66. 18 and 21 and 65 (1874)
67. 66 not 59 (135)

Interface: Science Citation Index via Web of Science

Date range searched: no restriction.

Search date: 17 March 2016.

Records identified: 113.

Search strategy

Set	Results	
#17	113	#16 not #13
		Indexes=SCI-EXPANDED Timespan=All years
#16	2987	#15 AND #9
		Indexes=SCI-EXPANDED Timespan=All years
#15	257,021	#14 OR #11 OR #10
		Indexes=SCI-EXPANDED Timespan=All years
#14	45,206	TOPIC: (intellectual* NEAR/2 (disabilit* or disabled or deficit* or handicap* or retard*)) *OR* TOPIC: (mental* NEAR/2 (disabilit* or disabled or deficit* or handicap* or retard*)) *OR* TOPIC: (learning NEAR/2 (disability or disabled or difficult*))
		Indexes=SCI-EXPANDED Timespan=All years
#13	2874	#12 AND #9
		Indexes=SCI-EXPANDED Timespan=All years
#12	227,721	#11 OR #10
		Indexes=SCI-EXPANDED Timespan=All years
#11	31,704	TOPIC: (developmental NEAR/2 (disabilit* or delay*)) *OR* TOPIC: (neurodisabilit*) *OR* TOPIC: (neurodevelopment* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) *OR* TOPIC: (neuromotor* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) *OR* TOPIC: (neuropsychiatric* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*)) *OR* TOPIC: (neuropsychol* NEAR/3 (delay* or disabilit* or disease* or disorder* or dysfunction*))
		Indexes=SCI-EXPANDED Timespan=All years

Set	Results	
#10	205,587	TOPIC: (ADHD or "attention deficit") *OR* TOPIC: ("angelman syndrome") *OR* TOPIC: (autism or autistic or asperger*) *OR* TOPIC: ("cerebral palsy")*OR* TOPIC: ("conduct disorder*") *OR* TOPIC: (epilepsy or epilepti) *OR* TOPIC: ("Down* syndrome") *OR* TOPIC: ("Fragile x syndrome") *OR* TOPIC: ("Prader Willi Syndrome") *OR* TOPIC: ("Rett syndrome") *OR* TOPIC: ("Smith-Magenis syndrome") *OR* TOPIC: ("Williams syndrome") *Indexes=SCI-EXPANDED Timespan=All years*
#9	12,558	#8 AND #7 *Indexes=SCI-EXPANDED Timespan=All years*
#8	1,509,601	TOPIC: (adolescen* or baby or babies or child or children or boy or boys or girl or girls or infant* or infanc* or juvenile* or paediatric or pediatric or preschooler* or schoolboy* or schoolgirl* or schoolchild* or teens or teenager* or toddler* or youth or youths or "young people" or "young person*") *Indexes=SCI-EXPANDED Timespan=All years*
#7	76,873	#6 OR #5 OR #4 OR #3 OR #2 OR #1 *Indexes=SCI-EXPANDED Timespan=All years*
#6	36,970	TS=(sleep* NEAR/3 (initiat* or pattern* or routine* or practice* or maintain* or intervention* or schedule*)) OR TS=(sleepless* or insomnia* or parasomnia* or "night terror*" or nightterror* or "night mare*" or nightmare*) OR TS=(sleep-wak* or night-wak*) OR TS=(sleepwalk* or sleep-walk* or "sleep walk*" or somnambulism) OR TS=(narcolepsy or "nocturnal hyperkinesia") *Indexes=SCI-EXPANDED Timespan=All years*
#5	48,375	TS=(nocturnal* NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(nocturnal* NEAR/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*)) OR TS=(sleep* NEAR/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*)) OR TS=(sleep* NEAR/3 (dysfunction* or disorder or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) *Indexes=SCI-EXPANDED Timespan=All years*
#4	3897	TS=("bed time*" NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(bedtime* NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(night* NEAR/3 (dysfunction* or disorder* or difficult* or disrupt* or disturb* or delay* or problem*)) OR TS=(night* NEAR/3 (settle* or settling or wake* or awake or wakeful* or waking* or awaking* or awakening* or wakening*)) *Indexes=SCI-EXPANDED Timespan=All years*
#3	4952	TS=(narcolepsy) *Indexes=SCI-EXPANDED Timespan=All years*
#2	299	TS=(somnambulism) *Indexes=SCI-EXPANDED Timespan=All years*
#1	33,931	TS=(sleep disorder*) *Indexes=SCI-EXPANDED Timespan=All years*

Applied Social Sciences Index and Abstract via Proquest

Date range searched: no restriction.

Search date: 17 March 2016.

Records identified: 12.

Search strategy

1. ((bed time or bedtime$) adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (2)
2. ((bed time$ or bedtime$) adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule$)).ti,ab. (12)
3. (night$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (33)
4. (night$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (44)
5. (nocturnal$ adj3 (dysfunction$ or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (4)
6. (nocturnal$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (7)
7. (sleep$ adj3 (settle$1 or settling or wake$1 or awake or wakeful$ or waking$ or awaking$ or awakening$ or wakening$)).ti,ab. (67)
8. (sleep$ adj3 (dysfunction$ or disorder or disorder$ or difficult$ or disrupt$ or disturb$ or delay$ or problem$)).ti,ab. (527)
9. (sleep$ adj3 (initiat$ or pattern$ or routine$ or practice$ or maintain$ or intervention$ or schedule$)).ti,ab. (260)
10. (sleepless$ or insomnia$ or parasomnia$ or night terror$ or nightterror$ or night mare$ or nightmare$).ti,ab. (258)
11. (sleep-wak$ or night-wak$).ti,ab. (41)
12. (sleepwalk$ or sleep-walk$ or sleep walk$ or somnambulism).ti,ab. (13)
13. (narcolepsy or nocturnal hyperkinesia).ti,ab. (5)
14. (ADHD or attention deficit).ti,ab. (941)
15. angelman syndrome.ti,ab. (8)
16. (autism or autistic or asperger$).ti,ab. (2638)
17. cerebral palsy.ti,ab. (344)
18. conduct disorder$.ti,ab. (431)
19. (epilepsy or epileptic).ti,ab. (292)
20. Down$ syndrome.ti,ab. (509)
21. Fragile x syndrome.ti,ab. (49)
22. Prader Willi Syndrome.ti,ab. (32)
23. Prader-Willi Syndrome.ti,ab. (32)
24. Rett syndrome.ti,ab. (20)
25. Smith-Magenis syndrome.ti,ab. (3)
26. Williams syndrome.ti,ab. (12)
27. (developmental adj2 (disabilit$ or delay$)).ti,ab. (590)
28. neurodisabilit$.ti,ab. (19)
29. (neurodevelopment$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (64)
30. (neuro-motor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (0)
31. (neuromotor$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (0)
32. (neuropsychiatric$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (32)
33. (neuropsychol$ adj3 (delay$ or disabilit$ or disease$ or disorder$ or dysfunction$)).ti,ab. (12)

34. or/1-13 (993)
35. or/14-33 (5507)
36. (adolescen$ or baby or babies or child or children or boy or boys or girl or girls or infant$ or infanc$ or juvenile$ or paediatric or pediatric or preschooler$ or schoolboy$ or schoolgirl$ or schoolchild$ or teens or teenager$ or toddler$ or youth or youths or young people or young person$).ti,ab. (139,979)
37. 34 and 35 and 36 (48)
38. (intellectual$ adj (disabilit$ or disabled or deficit$ or handicap$ or retard$)).ti,ab. (2135)
39. (mental$ adj (disabilit$ or disabled or deficit$ or handicap$ or retard$)).ti,ab. (1252)
40. (learning adj (disability or disabled or difficult$)).ti,ab. (5838)
41. 35 or 38 or 39 or 40 (13,791)
42. 34 and 36 and 41 (60)
43. 42 not 37 (12)

Appendix 3 The data extraction variables used for the study results

TABLE 30 Data extraction variables for study results (Microsoft Excel® 2010, Microsoft Corporation, Redmond, WA, USA)

Variable	Description
Study	Author, date
N_Treat	Number of participants in treatment arm
N_Treat2	Number of participants in second treatment arm (if applicable)
N_Treat3	Number of participants in third treatment arm (if applicable)
N_Control	Number of participants in control arm
Followup	Baseline '0'
Followup1	Final follow-up point for which subsequent data are being extracted (e.g. 12 weeks, 10 days)
Mean0_combined	Baseline mean score for crossover trials
Mean0_treat	Baseline mean score for treatment arm
LowerCI0_treat	Lower CI for baseline mean score for treatment arm
UpperCI0_treat	Upper CI for baseline mean score for treatment arm
SD0_combined	SD for baseline mean score for crossover trials
SD0_treat	SD for baseline mean score for treatment arm
SE0_treat	SE for baseline mean score for treatment arm
Median0_treat	Baseline median score for treatment arm
Min0_treat	Baseline minimum score for treatment arm
Max0_treat	Baseline maximum score for treatment arm
Range0_treat	Baseline range score for treatment arm
Q250_treat	Baseline lower quartile for treatment arm
Q750_treat	Baseline upper quartile for treatment arm
IQR0_treat	Baseline interquartile range for treatment arm
Mean0_plac	Baseline mean score for control/placebo arm
LowerCI0_ plac	Lower CI for baseline mean score for control/placebo arm
UpperCI0_ plac	Upper CI for baseline mean score for control/placebo arm
SD0_ plac	SD for baseline mean score for control/placebo arm
SE0_ plac	SE for baseline mean score for control/placebo arm
Median0_ plac	Baseline median score for control/placebo arm
Min0_ plac	Baseline minimum score for control/placebo arm
Max0_ plac	Baseline maximum score for control/placebo arm
Range0_ plac	Baseline range score for control/placebo arm
Q250_ plac	Baseline lower quartile for control/placebo arm

continued

TABLE 30 Data extraction variables for study results (Microsoft Excel® 2010, Microsoft Corporation, Redmond, WA, USA) (*continued*)

Variable	Description
Q750_ plac	Baseline upper quartile for control/placebo arm
IQR0_ plac	Baseline interquartile range for control/placebo arm
Mean1_treat	Follow-up mean score for treatment arm
LowerCI1_treat	Lower CI for follow-up mean score for treatment arm
UpperCI1_treat	Upper CI for follow-up mean score for treatment arm
SD1_treat	SD for follow-up mean score for treatment arm
SE1_treat	SE for follow-up mean score for treatment arm
Median1_treat	Follow-up mean median score for treatment arm
Min1_treat	Follow-up mean minimum score for treatment arm
Max1_treat	Follow-up mean maximum score for treatment arm
Range1_treat	Follow-up mean range score for treatment arm
Q251_treat	Follow-up mean lower quartile for treatment arm
Q751_treat	Follow-up mean upper quartile for treatment arm
IQR1_treat	Follow-up mean interquartile range for treatment arm
Mean1_ plac	Follow-up mean score for control/placebo arm
LowerCI1_ plac	Lower CI for follow-up mean score for control/placebo arm
UpperCI1_ plac	Upper CI for follow-up mean score for control/placebo arm
SD1_ plac	SD for follow-up mean score for control/placebo arm
SE1_ plac	SE for follow-up mean score for control/placebo arm
Median1_ plac	Follow-up mean median score for control/placebo arm
Min1_ plac	Follow-up mean minimum score for control/placebo arm
Max1_ plac	Follow-up mean maximum score for control/placebo arm
Range1_ plac	Follow-up mean range score for control/placebo arm
Q251_ plac	Follow-up mean lower quartile for control/placebo arm
Q751_ plac	Follow-up mean upper quartile for control/placebo arm
IQR1_ plac	Follow-up mean interquartile range for control/placebo arm
TimeEffect_F	ANOVA *f*-value for time effect
TimeEffect_P	ANOVA *p*-value for time effect
GroupEffect_F	ANOVA *f*-value for group effect
GroupEffect_P	ANOVA *p*-value for group effect
GroupTimeEffect_F	ANOVA *f*-value for interaction effect
GroupTimeEffect_P	ANOVA *p*-value for interaction effect
ChangeBL1_Mean_Treat	Mean change from baseline to follow-up for treatment arm
ChangeBL1_SD_Treat	SD of mean change from baseline to follow-up for treatment arm
ChangeBL1_SE_Treat	SE of mean change from baseline to follow-up for treatment arm
ChangeBL1_range_Treat	Range score in change from baseline to follow-up for treatment arm
ChangeBL1_LowerCI_Treat	Lower CI in change value from baseline to follow-up for treatment arm

TABLE 30 Data extraction variables for study results (Microsoft Excel® 2010, Microsoft Corporation, Redmond, WA, USA) (*continued*)

Variable	Description
ChangeBL1_UpperCI_Treat	Upper CI in change value from baseline to follow-up for treatment arm
ChangeBL1_P_Treat	*p*-value for change from baseline to follow-up for treatment arm
ChangeBL1_%_Treat	Percentage change from baseline to follow-up for treatment arm
ChangeBL1_Mean_plac	Mean change from baseline to follow-up for control/placebo arm
ChangeBL1_SD_ plac	SD of mean change from baseline to follow-up for control/placebo arm
ChangeBL1_SE_ plac	SE of mean change from baseline to follow-up for control/placebo arm
ChangeBL1_range_ plac	Range score in change from baseline to follow-up for control/placebo arm
ChangeBL1_LowerCI_ plac	Lower CI in change value from baseline to follow-up for control/placebo arm
ChangeBL1_UpperCI_ plac	Upper CI in change value from baseline to follow-up for control/placebo arm
ChangeBL1_P_ plac	*p*-value for change from baseline to follow-up for control/placebo arm
ChangeBL1_%_ plac	Percentage change from baseline to follow-up for control/placebo arm
Diff_Treat_Plac	Raw value/score of the difference between the treatment and control/placebo arm
Diff_Treat_Plac_mean	MD between the treatment and control/placebo arm
Diff_Treat_Plac_SD	SD of the MD between the treatment and control/placebo arm
Diff_Treat_Plac_test_statistic (DF)	Test statistic of the difference between the treatment and control/placebo arm
Diff_Treat_Plac_ES	Effect size of the difference between the treatment and control/placebo arm
Diff_Treat_Plac_P	*p*-value of the difference between the treatment and control/placebo arm
CarryOverEffect_P	Carry-over effect for crossover trials
Period_effect_P	Period effect for crossover trials
Period_T-P_Mean	Mean period effect in order of treatment to control/placebo for crossover trials
Period_T-P_SD	SD of the mean period effect in order of treatment to control/placebo for crossover trials
Period_P-T_Mean	Mean period effect in order of control/placebo to treatment for crossover trials
Period_P-T_SD	SD of the mean period effect in order of control/placebo to treatment for crossover trials
Period_diff_P	*p*-value for period effect
DiffChange_Mean	Difference in mean change from baseline to follow-up between the treatment and control/placebo arm
DiffChange_SD	SD of the difference in mean change from baseline to follow-up between the treatment and control/placebo arm
DiffChange_SE	SE of the difference in mean change from baseline to follow-up between the treatment and control/placebo arm
DiffChange_LowerCI	Lower CI of the difference in mean change from baseline to follow-up between the treatment and control/placebo arm
DiffChange_UpperCI	Upper CI of the difference in mean change from baseline to follow-up between the treatment and control/placebo arm
DiffChange_P	*p*-value of the difference in mean change from baseline to follow-up between the treatment and control/placebo arm
DiffChange_PES	Effect size of the difference in mean change from baseline to follow-up between the treatment and control/placebo arm

continued

TABLE 30 Data extraction variables for study results (Microsoft Excel® 2010, Microsoft Corporation, Redmond, WA, USA) (*continued*)

Variable	Description
AdjustedDiff_Mean	Adjusted difference in mean change from baseline to follow-up between the treatment and control/placebo arm
AdjustedDiff_SD	Adjusted SD of the difference in mean change from baseline to follow-up between the treatment and control/placebo arm
AdjustedDiff_SE	Adjusted SE of the difference in mean change from baseline to follow-up between the treatment and control/placebo arm
AdjustedDiff_LowerCI	Adjusted lower CI of the difference in mean change from baseline to follow-up between the treatment and control/placebo arm
AdjustedDiff_UpperCI	Adjusted upper CI of the difference in mean change from baseline to follow-up between the treatment and control/placebo arm
AdjustedDiff_P	*p*-value of the adjusted difference in mean change from baseline to follow-up between the treatment and control/placebo arm
Notes	Any additional notes of relevance

ANOVA, analysis of variance.

Note

Each variable was listed as the column heading in a spreadsheet, with data for each study extracted on each row underneath. One spreadsheet was used for each outcome reported.

Appendix 4 List of papers excluded after full-text review and reasons for exclusions

TABLE 31 List of papers excluded after full-text review and reasons for exclusion

Reference	Reason for exclusion
Records identified from trial registeries	
ISRCTN05534585, The use of MElatonin in children with Neuro-developmental Disorders and impaired Sleep; a randomised, double-blind, placebo-controlled, parallel study	Already included as Appleton *et al.*[48]
ISRCTN92655217, Snuggledown – Use of sensory blankets for children with autistic spectrum disorder	Already included as Gringras *et al.*[36]
NCT01322022, Treatment of sleep disturbances in young children with autism. 2009	Already included as Johnson *et al.*[107]
ISRCTN84194243, Development of effective primary care treatment of severe sleep disorders in children with a learning disability; a randomised controlled trial	Already included as Montgomery *et al.*[49]
ISRCTN77884120, Melatonin treatment for sleep problems in children with autism: a randomised controlled crossover trial	Already included as Wright *et al.*[106]
2007-004664-46, Influence of methylphenidate on sleep and circadian rhythm in children with Attention-Deficit/Hyperactivity Disorder (ADHD) – MELMET	Intervention
2011-003313-42, Agomelatine efficacy of the drug to improve sleep problems in autistic people	Intervention
ISRCTN31542578, A blinded randomised cross-over study of the effect of melatonin treatment of sleep disturbances on hypothalamic–pituitary-gonadal axis and leptin in pubertal children	Intervention
NCT00393042, Sleep and tolerability study: comparing the effects of Adderall XR and Focalin XR	Intervention
NCT00695136, The effect of Donepezil [Aricept (Registered Trademark)] on REM sleep in children with autism	Intervention
NCT00745030, Efficacy and tolerability of Ramelteon in patients with rapid eye movement (REM) behavior disorder and Parkinsonism	Intervention
NCT00807222, Effect of Vyvanse on sleep in children aged 6–12 years with attention deficit hyperactivity disorder (ADHD)	Intervention
NCT00989950, Study of the effect of individualizing Daytrana wear-times on sleep in children with ADHD	Intervention
NCT01156051, Effect of Guanfacine extended-release on attention deficit hyperactivity disorder (ADHD)-associated insomnia	Intervention
NCT01887132, A trial of the drug Donepezil for sleep enhancement and behavioral change in children with autism	Intervention
NCT02231008, Evaluating the effects of Tasimelteon vs Placebo on sleep disturbances in SMS	Intervention
NCT02487082, Pilot study of sleep therapy and biomarkers in children with autism spectrum disorders	Intervention
NCT02638168, Effects of evening dose of immediate release methylphenidate on sleep in children with ADHD	Intervention
NCT00152750, Study of Clonidine on sleep architecture in children with Tourette's Syndrome (TS) and comorbid ADHD	Outcome

continued

TABLE 31 List of papers excluded after full-text review and reasons for exclusion (*continued*)

Reference	Reason for exclusion
NCT01508793, Enhancing sleep duration: effects on children's eating and activity behaviors	Outcome
NCT01903681, Assessment of the pharmacokinetics of Circadin® in children with neurodevelopmental disorders and sleep disturbances	Outcome
NCT02132273, Use of an educational story to prepare children with developmental disabilities for sleep study	Outcome
IRCT2015062222865N1, The effect of aromatherapy with Rosa Damascena on sleep quality of children	Population
NCT00005753, Pharmacological and behavioral treatment of insomnia	Population
NCT00133055, Parenting matters: helping parents with young children	Population
NCT00877162, The rocky sleep study	Population
NCT02195401, The effects of a clean room sleeping environment on elemental and chemical concentrations in children with autism	Population
NCT02398214, A sleep hygiene-based intervention program for infants and toddlers	Population
NCT02648568, Does hypnosis improve severe sleepwalking?	Population
NTR4045, Effects of melatonin treatment, light therapy, and sleep improvement in children with sleep onset problems	Population
NCT00691080, Understanding sleep problems in children with autism spectrum disorder	Study design
Records identified from databases	
Adams JB, Holloway C. Pilot study of a moderate dose multivitamin/mineral supplement for children with autistic spectrum disorder. *J Altern Complement Med* 2004;**10**:1033–9	Intervention
Ahmann PA, Theye FW, Berg R, Linquist AJ, Van Erem AJ, Campbell LR. Placebo-controlled evaluation of amphetamine mixture-dextroamphetamine salts and amphetamine salts (Adderall): efficacy rate and side effects. *Pediatrics* 2001;**107**:E10	Intervention
Allen KD, Kuhn BR, DeHaai KA, Wallace DP. Evaluation of a behavioral treatment package to reduce sleep problems in children with Angelman Syndrome. *Res Dev Disabil* 2013;**34**:676–86	Study design (before-and-after study with ≤ 10 participants)
Ashkenasi A. Effect of transdermal methylphenidate wear times on sleep in children with attention deficit hyperactivity disorder. *Pediatr Neurol* 2011;**45**:381–6	Intervention
Askenasi A. O251. Effect of transdermal methylphenidate wear times on sleep in children with ADHD. 22nd meeting of the European Neurological Society, 9–12 June 2012, Prague, Czech Republic. *J Neurol* 2012;**259**:S33–34	Intervention
Barlow J, Cullen L. Coming together through touch: the experiences of parents of children with disabilities learning the principles of massage. *Early Child Dev Care* 2000;**161**:93–106	Intervention
Barlow J, Powell L, Cheshire A. The Training and Support Programme (involving basic massage) for parents of children with cerebral palsy: an implementation study. *J Bodyw Mov Ther* 2007;**11**:44–53	Intervention
Becke SP, Froehlich TE, Epstein JN. Effects of methylphenidate on sleep functioning in children with attention-deficit/hyperactivity disorder. *J Dev Behav Pediatr* 2016;**37**:395–404	Intervention
Blumer JL, Findling RL, Shih WJ, Soubrane C, Reed MD. Controlled clinical trial of zolpidem for the treatment of insomnia associated with attention-deficit/hyperactivity disorder in children 6 to 17 years of age. *Pediatrics* 2009;**123**:e770–6	Intervention
Bovet-du Bois N. [Neuroleptic treatment in child psychiatry: analysis of a series of 100 cases treated with thioridazine.] *Schweiz Rundsch Med Prax* 1973;**62**:1556–64	Intervention
Braam W, van Geijlswijk I, Keijzer H, Smits MG, Didden R, Curfs LM. Loss of response to melatonin treatment is associated with slow melatonin metabolism. *J Intellect Disabil Res* 2010;**54**:547–55	Intervention

TABLE 31 List of papers excluded after full-text review and reasons for exclusion (*continued*)

Reference	Reason for exclusion
Brady A, Mpetha K, Humphreys S, Carney AM. Developing a sleep service for children with learning disabilities or autistic spectrum disorders aged 0–5: setting up the service and lessons from practice. *Clinical Psychology Forum* 2011;**222**:31–5	Intervention
Briault M, Labrosse M, Verreault M, Berthiaume C, Lageix P, Turgeon L, Godbout R. Sleep in children with attention deficit disorders: effects of comorbid anxiety and response to treatment. 23rd Annual Meeting of the Associated-Professional-Sleep-Societies, 6–11 June 2009, Seattle, WA, USA. *Sleep* 2009;**32**:A65	Intervention
Buckley AW, Sassower K, Rodriguez AJ, Jennison K, Wingert K, Buckley J, *et al.* An open label trial of donepezil for enhancement of rapid eye movement sleep in young children with autism spectrum disorders. *J Child Adolesc Psychopharmacol* 2011;**21**:353–7	Intervention
Bussing R, Mason D, Garvan CW, Gurnani T, Koro-Ljungberg M, Noguchi K, Albarracin D. Willingness to use ADHD self-management: mixed methods study of perceptions by adolescents and parents. *J Child Fam Stud* 2016;**25**:562–73	Intervention
Campbell M, Perry R, Polonsky BB, Deutsch SI, Palij M, Lukashok D. An open study of fenfluramine in hospitalized young autistic children. *J Autism Dev Disord* 1986;**16**:495–506	Intervention
Campbell M, Deutsch SI, Perry R, Wolsky BB, Palij M. Short-term efficacy and safety of fenfluramine in hospitalized preschool-age autistic children: an open study. *Psychopharmacol Bull* 1986;**22**:141–7	Intervention
Campbell M, Small AM, Palij M, Perry R, Polonsky BB, Lukashok D, Anderson LT. The efficacy and safety of fenfluramine in autistic children: preliminary analysis of a double-blind study. *Psychopharmacol Bull* 1987;**23**:123–7	Intervention
Chen XQ, Zhang WN, Yang ZX, Zhao M, Cai FC, Huang SP, *et al.* Efficacy of levetiracetam in electrical status epilepticus during sleep of children: a multicenter experience. *Pediatr Neurol* 2014;**50**:243–9	Intervention
Cheng Song J, Hiscock H, Scibberras E, Schuster T. Behavioural sleep problems in children with ADHD: cross–sectional associations with parenting and sleep hygiene. *Sleep Biologic Rhythms* 2015;**13**(Suppl. 1):1–98	Intervention
Chung I, Han CH, Kim HW, Cho SC. P.7.c.008 The effect of prolonged-release methylphenidate on the sleep of children with attention-deficit/hyperactivity disorders. *Eur Neuropsychopharmacol* 2010;**20**(Suppl. 3):624–5	Intervention
Kim HW, Yoon IY, Cho SC, Kim BN, Chung S, Lee H, *et al.* The effect of OROS methylphenidate on the sleep of children with attention-deficit/hyperactivity disorder. *Int J Neuropsychopharmacol* 2010;**13**:184	Intervention
Clonidine for treatment of attention-deficit/hyperactivity disorder. *Med Lett Drugs Ther* 1996;**38**:109–10	Intervention
Cocchi R. Drug therapies for sleep troubles, hyperactivity and aggression in young adult autistics. *Ital J Intellective Impairment* 1995;**8**:169–74	Intervention
Cohen-Zion M. Sleep and circadian rhythms in children with attention deficit-hyperactivity disorder: before and after stimulant treatment. San Diego, CA: University of California, San Diego and San Diego State University; 2005	Intervention
De Leersnyder H, de Blois M-C, Vekemans M, Sidi D, Villain E, Kindermans C, Munnich A. β1-adrenergic antagonists improve sleep and behavioural disturbances in a circadian disorder, Smith–Magenis syndrome. *J Med Genet* 2001;**38**:586–90	Intervention
DeLeon IG, Fisher WW, Marhefka JM. Decreasing self-injurious behaviour associated with awakening in a child with autism and developmental delays. *Behav Interv* 2004;**19**:111–19	Intervention
Duncan B, Barton L, Edmonds D, Blashill BM. Parental perceptions of the therapeutic effect from osteopathic manipulation or acupuncture in children with spastic cerebral palsy. *Clin Pediatr* 2004;**43**:349–53	Intervention
Ediberidze T, Maisuradze L, Kasradze S. Influence of anticonvulsants on sleep difficulties in children with epileptic seizures of genetic aetiology (ESGE)-a questionnaire based study. *J Sleep Res* 2016;**25**:229	Intervention

continued

TABLE 31 List of papers excluded after full-text review and reasons for exclusion (*continued*)

Reference	Reason for exclusion
Rugino T. Effect of Guanfacine Extended-Release on Attention Deficit Hyperactivity Disorder (ADHD)-Associated Insomnia. Children's Specialised Hospital; 2010. URL: https://clinicaltrials.gov/ct2/show/NCT01156051 (accessed 26 July 2018)	Intervention
Efron D, Lycett K, Sciberras E. Use of sleep medication in children with ADHD. *Sleep Med* 2014;**15**:472–5	Intervention
Elbhrawy S, *et al.* Polysomnographic based study of sleep in juvenile myoclonic epilepsy patients. *Epilepsia* 2016;**57**(Suppl. 2):43	Intervention
Esmaeli L, Abedi MR, Najafi MR, Aminjafari A, Afsar F, Moghtadaei M. Effectiveness of emotion regulation on anxiety, insomnia and social dysfunction of epileptic adolescent girls. Abstracts of 5th International Congress Of Child and Adolescent Psychiatry, 8–11 October 2012, Tehran, Iran. *Iran J Psychiatry* 2012;**1**:3	Intervention
Faber S, Zinn GM, Boggess A, Fahrenholz T, Kern JC, Kingston HM. A cleanroom sleeping environment's impact on markers of oxidative stress, immune dysregulation, and behavior in children with autism spectrum disorders. *BMC Complement Altern Med* 2015;**15**:71	Intervention
Feinberg I, Hibi S, Braun M, Cavness C, Westerman G, Small A. Sleep amphetamine effects in MBDS and normal subjects. *Arch Gen Psychiatry* 1974;**31**:723–31	Intervention
Fletcher F, Foster-Owens M, Conduit R, Cornish K. The impact of sleep hygiene on sleep onset issues in children with high functioning autism and Asperger syndrome. *Sleep Biol Rhythms* **11**:12	Intervention
Fletcher, Rinehart N, Conduit R, Rajaratnam S, Thomas D, Cornish K. Targeting sleep problems and anxiety in children with high functioning autism. *Sleep Biol Rhythms* 2012;**10**:63	Intervention
Gastfriend DR, Biederman J, Jellinek MS. Desipramine in the treatment of adolescents with attention deficit disorder. *Am J Psychiatry* 1984;**141**:906–8	Intervention
Gastfriend DR, Biederman J, Jellinek MS. Desipramine in the treatment of attention deficit disorder in adolescents. *Psychopharmacol Bull* 1985;**21**:144–5	Intervention
Giblin JM, Strobel AL. Effect of lisdexamfetamine dimesylate on sleep in children with ADHD. *J Atten Disord* 2011;**15**:491–8	Intervention
Goldman SE, Adkins KW, Calcutt MW, Carter MD, Goodpaster RL, Wang L, *et al.* Melatonin in children with autism spectrum disorders: endogenous and pharmacokinetic profiles in relation to sleep. *J Autism Dev Disord* 2014;**44**:2525–35	Intervention
Grubar JC, Gigli GL, Colognola RM, Ferri R, Musumeci SA, Bergonzi P. Sleep patterns of Down's syndrome children: effects of butoctamide hydrogen succinate (BAHS) administration. *Psychopharmacology* 1986;**90**:119–22	Intervention
Gruber R, Grizenko N, Schwartz G, Ben Amor L, Gauthier J, de Guzman R, Joober R. Sleep and COMT polymorphism in ADHD children: preliminary actigraphic data. *J Am Acad Child Adolesc Psychiatry* 2006;**45**:982–9	Intervention
Gupta M, Aneja S, Kohli K. Add-on melatonin improves quality of life in epileptic children on valproate monotherapy: a randomised, double-blind, placebo-controlled trial. *Epilepsy Behav* 2004;**5**:316–21	Intervention
Hagerman RJ, Riddle JE, Roberts LS, Breese K, Fulton M. Survey of the efficacy of clonidine in fragile X syndrome. *Dev Brain Dysfunc* 1995;**8**:336–44	Intervention
Hallböök T, Lundgren J, Köhler S, Blennow G, Strömblad LG, Rosén I. Beneficial effects on sleep of vagus nerve stimulation in children with therapy resistant epilepsy. *Eur J Paediatr Neurol* 2005;**9**:399–407	Intervention
Hallböök T, Lundgren J, Rosén I. Ketogenic diet improves sleep quality in children with therapy-resistant epilepsy. *Epilepsia* 2007;**48**:59–65	Intervention
Head TK. Evaluation of medication effects on academic performance, sleep, and core ADHD symptoms in children. Kalamazoo, MI: Western Michigan University; 2015	Intervention

TABLE 31 List of papers excluded after full-text review and reasons for exclusion (*continued*)

Reference	Reason for exclusion
Henderson JA, Barry TD, Bader SH, Jordan SS. The relation among sleep, routines, and externalising behaviour in children with an autism spectrum disorder. *Res Autism Spectr Disord* 2011;**5**:758–67	Intervention
Hewitt K. Behavioural approaches to sleeplessness. *Br J Learning Disabil* 1985;**13**:112–14	Study design
Honomichl RD, Goodlin-Jones BL, Burnham MM, Hansen RL, Anders TF. Secretin and sleep in children with autism. *Child Psychiatry Hum Dev* 2002;**33**:107–23	Intervention
Hoshino K. Effectiveness of low dose of L-dopamine in children with autism and attention-deficithyperactivity disorder. *Dev Med Child Neurol* 2012;**54**:5–212	Intervention
Hunt RD, Minderaa RB, Cohen DJ. Clonidine benefits children with attention deficit disorder and hyperactivity: report of a double-blind placebo-crossover therapeutic trial. *J Am Acad Child Psychiatry* 1985;**24**:617–29	Intervention
Hunt RD, Minderaa RB, Cohen DJ. The therapeutic effect of clonidine in attention deficit disorder with hyperactivity: a comparison with placebo and methylphenidate. *Psychopharmacol Bull* 1986;**22**:229–36	Intervention
Hvolby A. Does treatment of ADHD sleeping problems improve attention, hyperactivity and impulsiveness in children with attention deficit hyperactivity disorder? *Eur Child Adolesc Psychiatry* 2013;**22**:S87–323	Intervention
Laboni A, Gibbons J, Hamilton J, Narang I. Sleep characteristics of prepubescent children with Prader–Willi syndrome before and after growth hormone treatment. *Sleep* 2009;**32**:A113	Intervention
Jan JE, Connolly MB, Hamilton D, Freeman RD, Laudon M. Melatonin treatment of non-epileptic myoclonus in children. *Dev Med Child Neurol* 1999;**41**:255–9	Intervention
Kent JD, Joseph C, Blader HS, Koplewicz HA, Foley CA. Effects of late-afternoon methylphenidate administration on behaviour and sleep in attention-deficit hyperactivity disorder. *Pediatrics* 1995;**96**:320–5	Intervention
Kim HW, Yoon IY, Cho SC, Kim BN, Chung S, Lee H, *et al.* The effect of OROS methylphenidate on the sleep of children with attention-deficit/hyperactivity disorder. *Int Clin Psychopharmacol* 2010;**25**:107–15	Intervention
Konafel E, Lecendreuz M, Bouvard MP, Mouren-Simeoni MC. Effects of vesperal methylphenidate (MPH) administration on diurnal and nocturnal activity in ADHD children: an actigraphic study. *Amer Acad Sleep Med* 2001:A215	Intervention
Liboni F, Palagini L, Mandredi A, Tacchi A, Ricci F, Mauri M, *et al.* Effects of six-months methylphenidate treatment on sleep disturbances in children with attention-deficit/ hyperactivity disorder. A pilot study. JOURNAL OF, 2014;**23**:322–2	Intervention
Lyon MR, Kapoor MP, Juneja LR. The effects of L-theanine (Suntheanine) on objective sleep quality in boys with attention deficit hyperactivity disorder (ADHD): a randomised, double-blind, placebo-controlled clinical trial. *Altern Med Rev* 2011;**16**:348–54	Intervention
Malow BA, MacDonald LL, Fawkes DB, Alder ML, Katz T. Teaching children with autism spectrum disorder how to sleep better: a pilot educational program for parents. *Clin Pract Paediat Psychol* 2016;**4**:125	Study design (before-and-after study with ≤ 10 participants)
Malow BA, Katz T, Reynolds AM, Shui A, Carno M, Connolly HV, *et al.* Sleep difficulties and medications in children with autism spectrum disorders: a registry study. *Pediatrics* 2016;**137**(Suppl. 2):98–104	Intervention
Miyamoto A, Fukuda I, Tanaka H, Oka R, Araki A, Cho K. [Treatment with ramelteon for sleep disturbance in severely disabled children and young adults.] *No To Hattatsu* 2013;**45**:440–4	Intervention
Mostafavi S-A, Mohammadi MR, Hosseinzadeh P, Eshraghian MR, Akhondzadeh R, Hosseinzadeh-Attar MJ, *et al.* Dietary intake, growth and development of children with ADHD in a randomised clinical trial of Ritalin and Melatonin co-administration: Through circadian cycle modification or appetite enhancement? *Iran J Psychiatry* 2012;**7**:114–19	Intervention

continued

TABLE 31 List of papers excluded after full-text review and reasons for exclusion (*continued*)

Reference	Reason for exclusion
Nayak C, Sinha S, Ramachandraiah CT, Nagappa M, Thennarasu K, Taly AB, Satishchandra P. Differential improvement of the sleep quality among patients with juvenile myoclonic epilepsy with valproic acid: A longitudinal sleep questionnaire-based study. *Ann Indian Acad Neurol* 2015;**18**:403–7	Intervention
Long Y, Tan J, Nie Y, Lu Y, Mei X, Tu C. Hyperbaric oxygen therapy is safe and effective for the treatment of sleep disorders in children with cerebral palsy. *Neurol Res* 2017;**39**:239–47	Intervention
Ornitz EM, Forsythe AB, de la Peña A. Effect of vestibular and auditory stimulation on the REMs of REM sleep in autistic children. *Arch Gen Psychiatry* 1973;**29**:786–91	Intervention
Owens J, Weiss M, Nordbrock E, Mattingly G, Wigal S, Greenhill LL, *et al.* Effect of aptensio XR (methylphenidate HCl extended-release) capsules on sleep in children with attention-deficit/hyperactivity disorder. *J Child Adolesc Psychopharmacol* 2016;**26**:873–81	Intervention
Petre-Quadens O, De Greef A. Effects of 5-HTP on sleep in Mongol children. Preliminary results. *J Neurol Sci* 1971;**13**:115–19	Intervention
Larsen S, Harrington K, Hicks S. The LENS (Low Energy Neurofeedback System): a clinical outcomes study on one hundred patients at Stone Mountain Centre, New York. *J Neurother* 2006;**10**:69–78	Intervention
Ramstad K, Jahnsen R, Lofterod B, Skjeldal OH. Continuous intrathecal baclofen therapy in children with cerebral palsy – when does improvement emerge? *Acta Paediatr* 2010;**99**:1661–5	Intervention
Robinson AA, Malow BA. Gabapentin shows promise in treating refractory insomnia in children. *J Child Neurol* 2013;**28**:1618–21	Intervention
Robinson AM, Richdale AL. Sleep problems in children with an intellectual disability: parental perceptions of sleep problems, and views of treatment effectiveness. *Child Care Health Dev* 2004;**30**:139–50	Intervention
Rogozea R, Florea-Ciocoiu V. Orienting reaction in patients with night terrors. *Biol Psychiatry* 1985;**20**:894–905	Intervention
Rugino TA. Effect on primary sleep disorders when children with ADHD are administered Guanfacine extended release. *J Atten Disord* 2014;**22**:14–24	Intervention
Binay Safer V, Ozbudak Demir S, Ozkan E, Demircioglu Guneri F. Effects of botulinum toxin serotype A on sleep problems in children with cerebral palsy and on mothers sleep quality and depression. *Neurosciences* 2016;**21**:331–7	Intervention
Sangal RB, Owens J, Allen AJ, Sutton V, Schuh K, Kelsey D. Effects of atomoxetine and methylphenidate on sleep in children with ADHD. *Sleep* 2006;**29**:1573–85	Intervention
Sangal RB, Blumer JL, Lankford DA, Grinnell TA, Huang H. Eszopiclone for insomnia associated with attention-deficit/hyperactivity disorder. *Pediatrics* 2014;**134**:e1095–103	Intervention
Sangal RB, Owens J, Allen A, Kelsey D, Sutton V, Schuh K. Effects of atomoxetine and methylphenidate on sleep in children with attention-deficit/hyperactivity disorder. *Int J Neuropsychopharmacol* 2004;7	Intervention
Schelleman M, Richdale A. The relative and combined effects of a diet and a behavioural intervention for behaviour and sleep problems in children with significant challenging behaviours. *Sleep Biol Rhythms* 2010;**8**:A7	Intervention
Shah T, Tse A, Gill H, Wong I, Sutcliffe A, Gringras P, *et al.* Administration of melatonin mixed with soft food and liquids for children with neurodevelopmental difficulties. *Dev Med Child Neurol* 2008;**50**:845–9	Intervention
Sherwin I, Hooge JP. Comparative effectiveness of natural sleep and methohexital. Provocative tests in electroencephalography. *Neurology* 1973;**23**:973–6	Intervention
Smits MG. Clinical experiences of DLMO measurements in insomnia patients at the Gelderse Vallei Hospital Sleep Centre. *J Sleep Res* 2010;**19**:97	Intervention
Strawn JR, McReynolds D. An evidence-based approach to treating paediatric anxiety disorders. *Current Psychiatry* 2012;**11**:16–21	Intervention

TABLE 31 List of papers excluded after full-text review and reasons for exclusion (*continued*)

Reference	Reason for exclusion
Tai A, Horne R, Davey M, Nixon G. The effects of growth hormone on sleep parameters in children with Prader–Willi syndrome. *Sleep Biol Rhythms* 2009;**7**:A56	Intervention
Tajima S, Matsuzawa S, Takai K, Kawakami M, Nakashima M, Miike T. Poster presentations 1. *Sleep Biol Rhythms* 2011;**9**:254–342	Intervention
Tatsumi Y, Mohri I, Shimizu S, Tachibana M, Ohno Y, Taniike M. Daytime physical activity and sleep in pre-schoolers with developmental disorders. *J Paediatr Child Health* 2015;**51**:396–402	Intervention
Tilford JM, Payakachat N, Kuhlthau KA, Pyne JM, Kovacs E, Bellando J, *et al.* Treatment for sleep problems in children with autism and caregiver spillover effects. *J Autism Dev Disord* 2015;**45**:3613–23	Intervention
Tirosh E, Sadeh A, Munvez R, Lavie P. Effects of methylphenidate on sleep in children with attention-deficit hyperactivity disorder: an activity monitor study. *Am J Dis Child* 1993;**147**:1313–15	Intervention
Tizard B. A controlled study of all-night sleep in overactive imbecile children. *Am J Ment Defic* 1968;**73**:209–13	Intervention
Valdizan JR, Almarcegui C, Brualla J, Alejos MV, Chulilla JL, Dolz I. The influence of gabapentin on sleep in children with secondarily generalised partial epilepsy. *Rev Neurol* 1999;**29**:718–21	Intervention
De Weerd A, Van Den Bosschhe R. Sleep in very young children with Prader Willi syndrome before and during growth hormone substitution. *J Sleep Res* 2010;**19**:29	Intervention
Van der Heijden KB, Smits MG, Gunning WB. Sleep hygiene and actigraphically evaluated sleep characteristics in children with ADHD and chronic sleep onset insomnia. *J Sleep Res* 2006;**15**:55–62	Intervention
Verrillo E, Bruni O, Franco P, Ferri R, Thiriez G, Pavone M, *et al.* Analysis of NREM sleep in children with Prader–Willi syndrome and the effect of growth hormone treatment. *Sleep Med* 2009;**10**:646–50	Intervention
Verrillo E, Bizzarri C, Bruni O, Ferri R, Pavone M, Cappa M, Cutrera R. Effects of replacement therapy on sleep architecture in children with growth hormone deficiency. *Sleep Med* 2012;**13**:496–502	Intervention
Wachob D, Lorenzi DG. Brief report: influence of physical activity on sleep quality in children with autism. *J Autism Dev Disord* 2015;**45**:2641–6	Intervention
Wang L, Liu Y. Behaviour improvement by diet and environmental interventions in brain injured children. *CRTER* 2007;**11**:5983–5	Intervention
Wehmeier PM, Dittman RW, Schacht A, Helsbery K, Lehmkuhl G. Morning and evening behaviour in children and adolescents treated with atomoxetine once daily for attention-deficit/hyperactivity disorder (ADHD): Findings from two 24-week, open-label studies. *Child Adolesc Psychiatry Ment Health* 2009;**3**:5	Intervention
Williams G, Sears L, Allard A. Parent perceptions of efficacy for strategies used to facilitate sleep in children with autism. *J Dev Phys Disabil* 2006;**18**:25–33	Intervention
Vol'f MSh, Prokudin VN, Iurkova IA. [The place of nitrazepam in the complex treatment of epilepsy.] *Zh Nevropatol Psikhiatr Im S S Korsakova* 1973;**73**:1714–18	Intervention
Wyatt K, Edwards V, Franck L, Britten N, Creanor S, Maddick A, Logan S. Cranial osteopathy for children with cerebral palsy: a randomised controlled trial. *Arch Dis Child* 2011;**96**:505–12	Intervention
Zametkin A, Rapoport JL, Murphy DL, Linnoila M, Ismond D. Treatment of hyperactive children with monoamine oxidase inhibitors. I. Clinical efficacy. *Arch Gen Psychiatry* 1985;**42**:962–6	Intervention
Zametkin AJ, Reeves JC, Webster L, Werry JS. Promethazine treatment of children with Attention Deficit Disorder with Hyperactivity – ineffective and unpleasant. *J Am Acad Child Psychiatry* 1986;**25**:854–6	Intervention

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TABLE 31 List of papers excluded after full-text review and reasons for exclusion (*continued*)

Reference	Reason for exclusion
Zandieh S, Khatwa U, Zarowski M, Kothare S. Utility of the maintenance of wakefulness test (MWT) in assessing treatment efficacy & optimising management in children with narcolepsy. 2012;**35**:A391	Intervention
Zhou JY, Tang XD, Huang LL, Zhong ZQ, Lei F, Zhou D. The acute effects of levetiracetam on nocturnal sleep and daytime sleepiness in patients with partial epilepsy. *J Clin Neurosci* 2012;**19**:956–60	Intervention
Beriault M, Turgeon L, Labrosse M, Berthiaume C, Verreault M, Berthiaume C, Godbout R. Comorbidity of ADHD and anxiety disorders in school-age children: impact on sleep and response to a cognitive-behavioural treatment. *J Atten Disord* 2015;**22**:22	Outcome
Borusiak P, Bast T, Kluger G, Weidenfeld A, Langer T, Jenke ACW, Wiegand G. A longitudinal, randomized, and prospective study of nocturnal monitoring in children and adolescents with epilepsy: Effects on quality of life and sleep. *Epilepsy Behav* 2016;**61**:192–8	Outcome
Bartlet L, Beaumont J. Treating the sleep disorders of children with disabilities and illness: a one-year project. *Clin Child Psychol Psychiatry* 1998;**3**:591–612	Population
Bazil CW, Dave J, Cole J, Stalvey J, Drake E. Pregabalin increases slow-wave sleep and may improve attention in patients with partial epilepsy and insomnia. *Epilepsy Behav* 2012;**23**:422–5	Population
Braam W, Didden R, Maas AP, Korzilius H, Smits MG, Curfs LM. Melatonin decreases daytime challenging behaviour in persons with intellectual disability and chronic insomnia. *J Intellect Disabil Res* 2010;**54**:52–9	Population
Braam W, Didden R, Smits M, Curfs L. Melatonin treatment in individuals with intellectual disability and chronic insomnia: a randomised placebo-controlled study. *J Intellect Disabil Res* 2008;**52**:256–64	Population
Brand S, Jossen S, Holsboer-Trachsler E, Pühse U, Gerber M. Impact of aerobic exercise on sleep and motor skills in children with autism spectrum disorders – a pilot study. *Neuropsychiatr Dis Treat* 2015;**11**:1911–20	Population
Burke RV, Kuhn BR, Peterson JL. Brief report: a 'storybook' ending to children's bedtime problems – the use of a rewarding social story to reduce bedtime resistance and frequent night waking. *J Pediatr Psychol* 2004;**29**:389–96	Population
Chang YS, Lin MH, Lee JH, Lee PL, Dai YS, Chu KH, *et al.*, Melatonin supplementation for children with atopic dermatitis and sleep disturbance a randomised clinical trial. *JAMA Pediatrics* 2016;**170**:35–42	Population
Coppola G, Iervolino G, Mastrosimone M, La Torre G, Ruiu F, Pascotto A. Melatonin in wake-sleep disorders in children, adolescents and young adults with mental retardation with or without epilepsy: a double-blind, cross-over, placebo-controlled trial. *Brain Dev* 2004;**26**:373–6	Population
Corkum P, Lingley-Pottie P, Davidson F, McGrath P, Chambers CT, Mullane J, *et al.* Better nights/better days – distance intervention for insomnia in school-aged children with/without ADHD: a randomised controlled trial. *J Paediatr Psychology* 2016;**41**:701–13	Population
Cronin S, Gose L, Gottschlich MM, Kagan RJ. Retrospective examination of the effectiveness of zolpidem for sleep in paediatric burn patients with a known history of attention deficit/hyperactivity disorder. *J Burn Care Res* 2012;**1**:S117	Population
Cronin SD, Gottschlich MM, Gose LM, Kagan RJ. Zolpidem and sleep in pediatric burn patients with attention deficit/hyperactivity disorder. *Pediatr Nurs* 2015;**41**:132–4, 140	Population
Bruin EJ, Bogels SM, Oort FJ, Meijer A. Improvements of adolescent psychopathology after insomnia treatment: results from a randomised controlled trial over 1 year. *J Sleep Res* 2016;**25**:91	Population
De Cock VC, Diene G, Molinas C, Masson VD, Kieffer I, Mimoun E, *et al.* Efficacy of modafinil on excessive daytime sleepiness in Prader–Willi syndrome. *Am J Med Genet A* 2011;**155A**:1552–7	Population

TABLE 31 List of papers excluded after full-text review and reasons for exclusion (*continued*)

Reference	Reason for exclusion
Einfeld SL, Smith E, McGregor IS, Steinbeck K, Taffe J, Rice LJ, *et al.* A double-blind randomized controlled trial of oxytocin nasal spray in Prader Willi syndrome. *Am J Med Genet A* 2014;**164A**:2232–9	Population
Freeman KA. Treating bedtime resistance with the bedtime pass: a systematic replication and component analysis with 3-year-olds. *J Appl Behav Anal* 2006;**39**:423–8	Population
Gigli GL, Grubar JC, Colognola RM, Amata MT, Pollicina C, Ferri R, *et al.* Butoctamide hydrogen succinate and intensive learning sessions: effects on night sleep of Down's syndrome patients. *Sleep* 1987;**10**:563–9	Population
Gupta M, Gupta YK, Aneja S, Kohli K. Effects of add-on melatonin on sleep in epileptic children on carbamazepine monotherapy: a randomised placebo controlled trial. *Sleep Biol Rhythms* 2004;**2**:215–19	Population
Gupta M, Aneja S, Kohli K. Add-on melatonin improves sleep behaviour in children with epilepsy: randomised, double-blind, placebo-controlled trial. *J Child Neurol* 2005;**20**:112–15	Population
Holsboer-Trachsler E. Aerobic exercise and skill training improved objective sleep and motor skills in children suffering from autism spectrum disorder (ASD) – a pilot study. *Biol Psychiatry* 2015;**77**:10010–1710	Population
Hvolby A, Bilenberg N. Use of Ball Blanket in attention-deficit/hyperactivity disorder sleeping problems. *Nord J Psychiatry* 2011;**65**:89–94	Population
Ishizaki A, Sugama M, Takeuchi N. [Usefulness of melatonin for developmental sleep and emotional/behaviour disorders–studies of melatonin trial on 50 patients with developmental disorders.] *No To Hattatsu* 1999;**31**:428–37	Population
Ivanenko A, Crabtree VM, Tauman R, Gozal D. Melatonin in children and adolescents with insomnia: a retrospective study. *Clin Pediatr* 2003;**42**:51–8	Population
Keshavarzi Z, Bajoghli H, Mohamadi MR, Salmanian M, Kirov R, Gerber M, *et al.* In a randomised case – control trial with 10-years olds suffering from attention deficit/ hyperactivity disorder (ADHD) sleep and psychological functioning improved during a 12-week sleep-training program. *World J Biol Psychiatry* 2014;**15**:609–19	Population
Ledet D, Aplin-Kalisz C, Filter M, Dycus P. A pilot study to assess a teaching intervention to improve sleep-wake disturbances in parents of children diagnosed with epilepsy. *J Neurosci Nurs* 2016;**48**:2–14	Population
McArthur AJ, Budden SS. Sleep dysfunction in Rett syndrome: a trial of exogenous melatonin treatment. *Dev Med Child Neurol* 1998;**40**:186–92	Population
Merrifield R. Evaluation of a health visitor-led sleep and behaviour clinic. *Community Pract* 2005;**78**:283–8	Population
Mila M, Cecilia A, Gracia A, Antonio M, Oscar T, Patricio P. Evaluation of oral iron supplementation in paediatric maintenance insomnia. *Sleep Med* 2013;**14**:e207–8	Population
Ming X, Gordon E, Kang N, Wagner GC. Use of clonidine in children with autism spectrum disorders. *Brain Dev* 2008;**30**:454–60	Population
Mohammadi MR, Mostafavi SA, Keshavarz SA, Eshraghian MR, Hosseinzadeh P, Hosseinzadeh-Attar MJ, *et al.* Melatonin effects in methylphenidate treated children with attention deficit hyperactivity disorder: a randomised double blind clinical trial. *Iran J Psychiatry* 2012;**7**:87–92	Population
Montgomery P, Burton JR, Sewell RP, Spreckelsen TF, Richardson AJ. Fatty acids and sleep in UK children: subjective and pilot objective sleep results from the DOLAB study – a randomised controlled trial. *J Sleep Res* 2014;**23**:364–88	Population
O'Callaghan FJ, Clarke AA, Hancock E, Hunt A, Osborne JP. Use of melatonin to treat sleep disorders in tuberous sclerosis. *Dev Med Child Neurol* 1999;**41**:123–6	Population
O'Connell A, Vannan K. Sleepwise: addressing sleep disturbance in young children with developmental delay. *Aust Occup Ther J* 2008;**55**:212–14	Population

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TABLE 31 List of papers excluded after full-text review and reasons for exclusion (*continued*)

Reference	Reason for exclusion
Paasch V, *et al.* Preparing children with autism spectrum disorders for overnight sleep studies: a case series. *Clin Pract Pediatr Psychol* 2016;**4**:153–63	Population
Pelsser LM, Frankena K, Buitelaar JK, Rommelse NN. Effects of food on physical and sleep complaints in children with ADHD: a randomised controlled pilot study. *Eur J Pediatr* 2010;**169**:1129–38	Population
Prince JB, Wilens TE, Biederman J, Spencer TJ, Wozniak JR. Clonidine for sleep disturbances associated with attention-deficit hyperactivity disorder: a systematic chart review of 62 cases. *J Am Acad Child Adolesc Psychiatry* 1996;**35**:599–605	Population
Ross C, Davies P, Whitehouse W. Melatonin treatment for sleep disorders in children with neurodevelopmental disorders: an observational study. *Dev Med Child Neurol* 2002;**44**:339–44	Population
Schlarb AA, Brandhorst I, Hautzinger M. [Mini-KiSS – a multimodal group therapy intervention for parents of young children with sleep disorders: a pilot study.] *Z Kinder Jugendpsychiatr Psychother* 2011;**39**:197–206	Population
Schroder C, Schmidt C, Kilic-huck U, Danion-Grilliat A, Bourgin P. Impact of long-term melatonin treatment for sleep disturbances in a child psychiatric population. *Eur Child Adolesc Psychiatry* 2015;**24**:1–303	Population
Silva LM, Scalock M, Ayres R, Bunse C, Budden S. Qigong massage treatment for sensory and self-regulation problems in young children with autism: a randomised controlled trial. *Am J Occupation Ther* 2009;**63**:423–32	Population
Simcock G. Sleep problems: assessing behavioural approaches. *Community Pract* 1999;**72**:128–30	Population
Smits MG, Nagtegaal EE, van der Heijden J, Coenen AM, Kerkhof GA. Melatonin for chronic sleep onset insomnia in children: a randomized placebo-controlled trial. *J Child Neurol* 2001;**16**:86–92	Population
Smits MG, van der Heijden KR, Meijer A, Coenen AM, Kerkhof GA. Melatonin improves health status and sleep in children with idiopathic chronic sleep-onset insomnia: a randomised placebo-controlled trial. *J Am Acad Child Adolesc Psychiatry* 2003;**42**:1286–93	Population
Stores R, Stores G. Evaluation of brief group-administered instruction for parents to prevent or minimise sleep problems in young children with Down syndrome. *J Appl Res Intellect Disabil* 2004;**17**:61–70	Population
Szeinberg A, Borodkin K, Dagan Y. Melatonin treatment in adolescents with delayed sleep phase syndrome. *Clin Pediatr* 2006;**45**:809–18	Population
Van Maanen A, Meijer AM, Smits MG, Oort FJ. Termination of short term melatonin treatment in children with delayed Dim Light Melatonin Onset: effects on sleep, health, behaviour problems, and parenting stress. *Sleep Med* 2011;**12**:875–9	Population
Ward F, Nanjappa M, Hinder SA, Roy M. Use of melatonin for sleep disturbance in a large intellectual disability psychiatry service. *Int J Developmental Disabil* 2015;**61**:182–7	Population
Williams TI. Evaluating effects of aromatherapy massage on sleep in children with autism: a pilot study. *Evid Based Complement Alternat Med* 2006;**3**:373–7	Population
Abdollahian E, Mohareri F. Evaluation of the effect of melatonin on improvement of sleep quality in children with attention deficit/hyperactivity disorder whom received ritalin. *Eur Psychiatry* 2015;**30**(Suppl. 1):584	Study design
Catherall C, Williams-Jones A. Managing sleep problems in children. *Learning Disability Practice* 2011;**14**:14–19	Study design
Andersen IM, Kaczmarska J, McGrew SG, Malow BA. Melatonin for insomnia in children with autism spectrum disorders. *J Child Neurol* 2008;**23**:482–5	Study design
Anonymous. Does melatonin improve sleep in children with neurodevelopmental disorders? *DTB* 2013;**51**:4	Study design
Anonymous. Melatonin for primary insomnia? *DTB* 2009;**47**:74–7	Study design

TABLE 31 List of papers excluded after full-text review and reasons for exclusion (*continued*)

Reference	Reason for exclusion
Anonymous. Melatonin improves sleep in children with ADHD and chronic insomnia: no effect found on behaviour, cognition or quality of life. *CABL* 2007;**23**:1–7	Study design
Anonymous. Neurodevelopmental disorders: No evidence for efficiency of weighted blankets in improving sleep in children with autism spectrum disorder. *Nat Rev Neurol* 2014;**10**:428	Study design
Arns M, Kenemans JL. Neurofeedback in ADHD and insomnia: vigilance stabilisation through sleep spindles and circadian networks. *Neurosci Biobehav Rev* 2014;**44**:183–94	Study design
Ayyash HF, Preece P, Morton R, Cortese S. Melatonin for sleep disturbance in children with neurodevelopmental disorders: prospective observational naturalistic study. *Expert Rev Neurother* 2015;**15**:711–17	Study design
Behavioural Interventions To Improve Sleep Disturbance in Typically Developing Children May be Effective for Children with Autism Spectrum Disorder. Clinician's Research Digest: Adult Populations. 2012;**30**:6	Study design
Bourdon GG. Treating insomnia in children with ADHD . . . 'towards a psychology of understanding: attention-deficit/hyperactivity disorder'. *JAAPA* 1998;**11**:76	Study design
Bouvier M, Claustrat B, Franco P. Melatonin treatment in autism spectrum disorders (ASD): preliminary results. *J Sleep Res* 2012;**236**	Study design
Brown LW. Looking beyond the polysomnograph in ADHD. *Sleep* 2006;**29**:745–6	Study design
Christodulu KV, Durand VM. Reducing bedtime disturbance and night waking using positive bedtime routines and sleep restriction. *Focus Autism Other Dev Disabli* 2004;**19**:130–9	Study design
Christodulu KV. Reducing bedtime disturbances and night waking using positive bedtime routines and sleep restriction. *Diss Abstr Int* 2000;**3**:130–9	Study design
Colville GA, Watters JP, Yule W, Bax M. Sleep problems in children with Sanfilippo syndrome. *Dev Med Child Neurol* 1996;**38**:538–44	Study design
Cortese S, Brown TE, Corkum P, Gruber R, O'Brien LM. First step in sleep problems in ADHD is promoting healthy sleep habits. *CABL Update* 2013;**15**:8	Study design
Damiani JM, Sweet BV, Sohoni P. Melatonin: an option for managing sleep disorders in children with autism spectrum disorder. *Am J Health Syst Pharm* 2014;**71**:95–101	Study design
De Leersnyder H, Bresson JL, de Blois MC, Souberbielle JC, Mogenet A, Delhotal-Landes B, *et al.* Beta 1-adrenergic antagonists and melatonin reset the clock and restore sleep in a circadian disorder, Smith–Magenis syndrome. *J Med Genet* 2003;**40**:74–8	Study design
De Leersnyder H, Zisapel N, Laudon M. Prolonged-release melatonin for children with neurodevelopmental disorders. *Pediatr Neurol* 2011;**45**:23–6	Study design
Didden R, Curfs LM, van Driel S, de Moor JM. Sleep problems in children and young adults with developmental disabilities: home-based functional assessment and treatment. *J Behav Ther Exp Psychiatry* 2002;**33**:49–58	Study design
Didden R, de Moor JMH, Curfs LMG, Behavioural treatment of sleep problems in three children with developmental disabilities. *BJDD* 2004;**50**:13–19	Study design
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Does melatonin improve sleep in children with neurodevelopmental disorders? *Drug Ther Bull* 2013;**51**:4	Study design
Durand VM, Christodulu KV. Description of a sleep-restriction program to reduce bedtime disturbances and night waking. *J Posit Behav Interv* 2004;**6**:83–91	Study design
Durand VM, Gernert-Dott P, Mapstone E. Treatment of sleep disorders in children with developmental disabilities. *Res Pract Persons Severe Disabl* 1996;**21**:114–22	Study design
Durand VM. Treating sleep terrors in children with autism. *J Posit Behav Interv* 2002;**4**:66–72	Study design

continued

TABLE 31 List of papers excluded after full-text review and reasons for exclusion (*continued*)

Reference	Reason for exclusion
Duwez M, Rouby A, Renou M, Chanoine S, Sabourdy C, Vercueil L, *et al.* Effectiveness and safety of oral melatonin premedication in sleep EEG in paediatrics. *Int J Clin Pharm* 2016;**38**:580–1	Study design
Efron D, Lycett K, Sciberras E. Response to: sleep medication in patients with attention-deficit/hyperactivity disorder. *Sleep Med* 2015;**16**:208	Study design
Elkhayat HA, Hassanein SM, Tomoum HY, Abd-Elhamid IA, Asaad T, Elwakkad AS. Melatonin and sleep-related problems in children with intractable epilepsy. *Pediatr Neurol* 2010;**42**:249–54	Study design
Esbensen AJ, Beebe DW, Byars KC, Hoffman EK. Use of sleep evaluations and treatments in children with Down syndrome. *J Dev Behav Pediatr* 2016;**37**:629–36	Study design
Espie CA, Wilson A. Improving sleep-wake schedules among people with mental handicaps: some preliminary case material. *Behav Cogn Psychother* 1993;**21**:51–5	Study design
Fauteck J, Schmidt H, Lerchl A, Kurlemann G, Wittkowski W. Melatonin in epilepsy: first results of replacement therapy and first clinical results. *Biol Signals Recept* 1999;**8**:105–10	Study design
Gee BM, Peterson T, Buck A, Lloyd K. Improving sleep quality using weighted blankets among young children with an autism spectrum disorder. *Int J Ther Rehabil* 2016;**23**:173–81	Study design
Giannotti F, Cortesi F, Cerquiglini A, Bernabei P. An open-label study of controlled-release melatonin in treatment of sleep disorders in children with autism. *J Autism Dev Disord* 2006;**36**:741–52	Study design
Giannotti F, Cortesi F, Antonella C, Bernabei P. Long-term melatonin treatment for sleep disorders in autistic children. A two-year follow-up study. *Amer Acad Sleep Med* 2004:94–4	Study design
Greydanus DE. Causes and management of sleep problems in children with developmental disabilities. *Child Care Health Dev* 2010;**36**:29	Study design
Gupta R, Hutchins J. Melatonin: a panacea for desperate parents? (Hype or truth). *Arch Dis Child* 2005;**90**:986–7	Study design
Hart-Santora D, Hart LL. Clonidine in attention deficit hyperactivity disorder. *Ann Pharmacother* 1992;**26**:37–9	Study design
Hätönen T, Kirveskari E, Heiskala H, Sainio K, Laakso ML, Santavuori P. Melatonin ineffective in neuronal ceroid lipofuscinosis patients with fragmented or normal motor activity rhythms recorded by wrist actigraphy. *Mol Genet Metab* 1999;**66**:401–6	Study design
Herrmann S. Counting sheep: sleep disorders in children with autism spectrum disorders. *J Pediatr Health Care* 2016;**30**:143–54	Study design
Hodoba D, Schmidt D. Biperiden for treatment of somnambulism in adolescents and adults with or without epilepsy: clinical observations. *Epilepsy Behav* 2012;**25**:517–28	Study design
Howlin P. A brief report on the elimination of long term sleeping problems in a 6-year-old autistic boy. *Behav Cogn Psychother* 1984;**12**:257–60	Study design
Hvolby A, Bilenberg N. Use of ball blanket in attention deficit hyperactivity disorder sleeping problems. *Eur Child Adolesc Psychiatry* 2011;**20**:7–223	Study design
Hylkema T, Vlaskamp C. Significant improvement in sleep in people with intellectual disabilities living in residential settings by non-pharmaceutical interventions. *J Intellect Disabil Res* 2009;**53**:695–703	Study design
Ingrassia A, Turk J. The use of clonidine for severe and intractable sleep problems in children with neurodevelopmental disorders – a case series. *Eur Child Adolesc Psychiatry* 2005;**14**:34–40	Study design
Itoh M, Hayashi M, Hasegawa T, Shimohira M, Kohyama J. Systemic growth hormone corrects sleep disturbance in Smith–Magenis syndrome. *Brain Dev* 2004;**26**:484–6	Study design
Jan JE, Espezel H. Melatonin treatment of chronic sleep disorders. *Dev Med Child Neurol* 1995;**37**:279–80	Study design

TABLE 31 List of papers excluded after full-text review and reasons for exclusion (*continued*)

Reference	Reason for exclusion
Jan JE, O'Donnell ME. Use of melatonin in the treatment of paediatric sleep disorders. *J Pineal Res* 1996;**21**:193–9	Study design
Jan JE, Espezel H, Goulden KJ. Melatonin in sleep disorders of children with neurodevelopmental disabilities. Melatonin in psychiatric and neoplastic disorders. *Progress Psychiatry* 1998;**55**:169–88	Study design
Jan JE, Freeman RD, Fast DK. Melatonin treatment of sleep–wake cycle disorders in children and adolescents. *Dev Med Child Neurol* 1999;**41**:491–500	Study design
Jan JE, Espezel H, Appleton RE. The treatment of sleep disorders with melatonin. *Dev Med Child Neurol* 1994;**36**:97–107	Study design
Jan MM. Melatonin for the treatment of handicapped children with severe sleep disorders. *Pediatr Neurol* 2000;**23**:229–32	Study design
Jin CS, Hanley GP, Beaulieu L. An individualized and comprehensive approach to treating sleep problems in young children. *J Appl Behav Anal* 2013;**46**:161–80	Study design
Kawabe K, Horiuchi F, Oka Y, Ueno S. The melatonin receptor agonist ramelteon effectively treats insomnia and behavioural symptoms in autistic disorder. *Case Rep Psychiatry* 2014;**2014**:561071	Study design
Knight R, Johnson M. Using a behavioural treatment package for sleep problems in children with autism spectrum disorders. *Child Fam Behav Ther* 2014;**36**:204–21	Study design
Konishi T, Masuko K, Naganuma Y, Hongou K, Yagi S. Flunitrazepam for sleep disturbance in children with intractable epilepsy. *Brain Dev* 1995;**17**:69–72	Study design
Lerchl A, Reiter RJ. Treatment of sleep disorders with melatonin. *BMJ* 2012;**345**:e6968	Study design
Malow B, Adkins KW, McGrew SG, Wang L, Goldman SE, Fawkes D, Burnette C. Melatonin for sleep in children with autism: a controlled trial examining dose, tolerability, and outcomes. *J Autism Dev Disord* 2012;**42**:1729–37	Study design
Malow BA, Adkins KW, McGrew SG, Wang L, Goldman SE, Fawkes D, Burnette C. Erratum to: Melatonin for sleep in children with autism: a controlled trial examining dose, tolerability, and outcomes. *J Autism Dev Disord* 2012;**42**:1738	Study design
Malow BA, Adkins KW, McGrew SG, Surdyka K, Goldman SE, Wofford D. Impact of supplemental melatonin on sleep and behaviour in children with autism spectrum disorders. 2009;**32**:A64	Study design
Malow BA, Adkins K, Goldman SE, McGrew SG, Burnette C, Wofford D, *et al.* Supplemental melatonin decreases sleep latency in children with autism. *Amer Acad Sleep Med* 2011:A267	Study design
Malow BA, Adkins KW, McGrew SG, Surdyka K, Wofford D. Supplemental melatonin improves sleep in children with autism spectrum disorders. *Ann Neurol* 2009;**66**:S31	Study design
McArthur I, Maisey J, Zuberi SM. Does slow release melatonin (SR) prevent recurrent night wakenings in children with neurodisability? *Amer Academy Sleep* 2003;**26**:A122–3	Study design
McArthur I, Maisey J, Zuberi SM. Impact of a nurse led sleep clinic on the use and evaluation of melatonin FR (Fast release) for sleep problems in children with neurodisability. *Amer Academy Sleep* 2003;**126**:A123	Study design
McLay LLK, France K. Empirical research evaluating non-traditional approaches to managing sleep problems in children with autism. *Dev Neurorehabil* 2014;**19**:123–34	Study design
Melatonin for insomnia in ADHD. Nurses' Drug Alert. *AJN* 2010;**34**:3–4	Study design
The Brown University Child and Adolescent Behaviour Letter (CABL) 2007;**23**:1–7. URL: https://onlinelibrary.wiley.com/doi/epdf/10.1002/cbl.20043 (accessed 26 July 2018)	Study design
Miyamoto A, Oki J, Takahashi S, Okuno A. Serum melatonin kinetics and long-term melatonin treatment for sleep disorders in Rett syndrome. *Brain Dev* 1999;**21**:59–62	Study design

continued

TABLE 31 List of papers excluded after full-text review and reasons for exclusion (*continued*)

Reference	Reason for exclusion
Moon EC, Corkum P, Smith IM. Case study: a case-series evaluation of a behavioural sleep intervention for three children with autism and primary insomnia. *J Pediatr Psychol* 2011;**36**:47–54	Study design
Moore P. The use of social stories in a psychology service for children with learning disabilities: a case study of a sleep problem. *Br J Learn Disabil* 2004;**32**:133–8	Study design
Mullane J, Corkum P. Case series: evaluation of a behavioral sleep intervention for three children with attention-deficit/hyperactivity disorder and dyssomnia. *J Atten Disord* 2006;**10**:217–27	Study design
Nakano T, Koyama E, Taniike M, Unase K. Application of an infrared sensor to home-monitoring of rest-activity patterns in a child with sleep disturbance. *Sleep Biol Rhythms* 2003;**1**:173–4	Study design
Narasingharao K, Pradhan B, Navaneetham J. Sleep Disorder, Gastrointestinal Problems and Behaviour Problems Seen in Autism Spectrum Disorder Children and Yoga as Therapy: A Descriptive Review. *J Clin Diagn Res* 2016;**10**:VE01–3	Study design
Niederhofer H, Staffen W, Mair A, Pittschieler K. Brief report: melatonin facilitates sleep in individuals with mental retardation and insomnia. *J Autism Dev Disord* 2003;**33**:469–72	Study design
Paavonen EJ, Nieminen-von Wendt T, Vanhala R, Aronen ET, von Wendt L. Effectiveness of melatonin in the treatment of sleep disturbances in children with Asperger disorder. *J Child Adolesc Psychopharmacol* 2003;**13**:83–95	Study design
Palm L, Blennow G, Wetterberg L. Long-term melatonin treatment in blind children and young adults with circadian sleep-wake disturbances. *Dev Med Child Neurol* 1997;**39**:319–25	Study design
Piazza CC, Fisher W. A faded bedtime with response cost protocol for treatment of multiple sleep problems in children. *J Appl Behav Anal* 1991;**24**:129–40	Study design
Piazza CC, Fisher WW. Bedtime fading in the treatment of pediatric insomnia. *J Behav Ther Exp Psychiatry* 1991;**22**:53–6	Study design
Piazza CC, Hagopian LP, Hughes CR, Fisher WW. Using chronotherapy to treat severe sleep problems: a case study. *Am J Ment Retard* 1998;**102**:358–66	Study design
Piazza CC, Fisher W, Moser H. Behavioral treatment of sleep dysfunction in patients with the Rett syndrome. *Brain Dev* 1991;**13**:232–7	Study design
Pillar G, Shahar E, Peled N, Ravid S, Lavie P, Etzioni A. Melatonin improves sleep-wake patterns in psychomotor retarded children. *Pediatr Neurol* 2000;**23**:225–8	Study design
Pillar G, Etzioni A, Shahar E, Lavie P. Melatonin treatment in an institutionalised child with psychomotor retardation and an irregular sleep-wake pattern. *Arch Dis Child* 1998;**79**:63–4	Study design
Reeve A, Miers S. Managing sleep problems in children with special needs. *Health Visit* 1994;**67**:230–1	Study design
Richdale AL. Sleep problems in autism: prevalence, cause, and intervention. *Dev Med Child Neurol* 1999;**41**:60–6	Study design
Richdale AL. Treatment of sleep problems in children with autism. *J Intellect Disabil Res* 2000;**44**:441–1	Study design
Ross C, Morris B, Whitehouse W. Melatonin treatment of sleep-wake cycle disorders in children and adolescents. *Dev Med Child Neurol* 1999;**41**:850	Study design
Rukovets O. Mending Children's sleep habits, improving family relations. *Neurol Today* 2012;**12**:37	Study design
Scott J. Melatonin an effective treatment for sleep problems in children with autism. *Foods Matter (USA)* 2009:15	Study design
Snowden S. Question 1. Does melatonin improve sleep pattern in children with attention deficit hyperactivity disorder? *Arch Dis Child* 2009;**94**:321–2	Study design

TABLE 31 List of papers excluded after full-text review and reasons for exclusion (*continued*)

Reference	Reason for exclusion
Stevinson C. Valerian may improve sleep of children with intellectual deficits. *Focus Altern Complement Ther* 2002;**7**:347–8	Study design
Summers JA, Lynch PS, Harris JC, Burke JC, Allison DB, Sandler L. A combined behavioural/pharmacological treatment of sleep–wake schedule disorder in Angelman syndrome. *J Dev Behav Pediatr* 1992;**13**:284–7	Study design
Thackeray EJ, Richdale AL. The behavioural treatment of sleep difficulties in children with an intellectual disability. *Behav Interv* 2002;**17**:211–31	Study design
Tjon Pian Gi CV, Broeren JP, Starreveld JS, Versteegh FG. Melatonin for treatment of sleeping disorders in children with attention deficit/hyperactivity disorder: a preliminary open label study. *Eur J Pediatr* 2003;**162**:554–5	Study design
Top tips. *Learning Disability Today*. 2013;**13**:20	Study design
Toussaint FS, Baverstock AC. An audit of melatonin use and parental satisfaction in a district general hospital. *Arch Dis Child* 2012;**97**(Suppl. 1):A128	Study design
Uberos J, Augustin-Morales MC, Molina Carballo A, Florido J, Narbona E, Munoz-Hoyos A. Normalisation of the sleep-wake pattern and melatonin and 6-sulphatoxy-melatonin levels after a therapeutic trial with melatonin in children with severe epilepsy. *J Pineal Res* 2011;**50**:192–6	Study design
Wassmer E, Carter PF, Quinn E, McLean N, Welsh G, Seri S, Whitehouse WP. Melatonin is useful for recording sleep EEGs: a prospective audit of outcome. *Dev Med Child Neurol* 2001;**43**:735–8	Study design
Waters F. Natural alternative offers hope on sleep disorders: the National Network for Learning Disability Nurses held its annual conference in Pontypridd last week. *Nurs Stand* 2004;**18**:9–10	Study design
Weiskop S, Matthews J, Richdale A. Treatment of sleep problems in a 5-year-old boy with autism using behavioural principles. *Autism* 2001;**5**:209–21	Study design
Wilens TE, Spencer TJ, Swanson JM, Connor DF, Cantwell D. Combining methylphenidate and clonidine: a clinically sound medication option. *J Am Acad Child Adolesc Psychiatry* 1999;**38**:614–19	Study design
Yamashita Y, Matsuishi T, Murakami Y, Kato H. Sleep disorder in Rett syndrome and melatonin treatment. *Brain Dev* 1999;**21**:570	Study design
Zhdanova IV, Wurtman RJ, Wagstaff J. Effects of a low dose of melatonin on sleep in children with Angelman syndrome. *J Pediatr Endocrinol Metab* 1999;**12**:57–67	Study design
Zolpidem not effective for ADHD-associated insomnia. *CABL Update* 2009;**11**:7	Study design
Zotter H, Kerbl R, Millner M, Kurz R. Methylphenidate and melatonin for sleep disorder with optic glioma. *J Am Acad Child Adolesc Psychiatry* 2001;**40**:992–3	Study design

Appendix 5 Study details for pharmacological interventions

TABLE 32 Study details for pharmacological interventions

Study details and study design	Trial treatments	Participant characteristics: number randomised, mean age (SD); % male, % diagnosis	Type of sleep disturbance/prior interventions	Risk of bias (low, unclear or high)
Melatonin vs. placebo *Parallel trials*				
Appleton *et al.*[48] Associated publications: Gringras *et al.*[108] and Appleton *et al.*[48,109] UK	Melatonin: capsules, starting dose of 0.5 mg, taken 45 minutes before bedtime for 12 weeks. Dose could be raised to 2, 6 and 12 mg in the first 4 weeks, then maintained Placebo: capsule-matching placebo, starting dose of 0.5 mg. Dose could be escalated through 2 mg and 6 mg to 12 mg in the first 4 weeks, then maintained	$N = 146$ Melatonin ($n = 70$): 106.0 months (34.8 months), range 44–181 months 70% male DD, 19%; DD and epilepsy, 11%; DD and ASD, 43%; DD, ASD and epilepsy, 0; DD and 'other', 27% Placebo ($n = 76$): 100.7 months (37.4 months), range 37–186 months 63% male DD, 9%; DD and epilepsy, 7%; DD and ASD, 39%; DD, ASD and epilepsy, 3%; DD and 'other', 38%	Sleep disturbance • Sleep initiation: not falling asleep within 1 hour of 'lights off' or 'snuggling down to sleep' in 3 out of 5 nights and/or < 6 hours • Sleep maintenance: continuous sleep in 3 out of 5 nights Prior interventions • Prior to randomisation, research nurses introduced eligible participants to a behaviour therapy advice booklet and children's sleep was monitored using parent-completed sleep diaries and actigraphy. Only children whose sleep problems persisted were randomised	Low
Cortesi *et al.*[110] Italy	Melatonin: controlled release, dose 3 mg, taken at 9 p.m. for 12 weeks CBT: 4 sessions, weekly individual sessions Melatonin and CBT: as above	$N = 160$ Melatonin ($n = 40$): 6.8 years (0.9 years) 82% male ASD, 100%	Sleep disturbance • Sleep initiation: mixed sleep onset • Sleep maintenance: maintenance insomnia Prior interventions	High

Study details and study design	Trial treatments	Participant characteristics: number randomised, mean age (SD); % male, % diagnosis	Type of sleep disturbance/prior interventions	Risk of bias (low, unclear or high)
	Placebo: identical placebo tablet (same appearance smell and flavour to active treatment), dose 3 mg, taken at 9 p.m. for 12 weeks	Melatonin and CBT ($n = 40$): 6.4 years (1.1 years) 80% male ASD, 100% CBT ($n = 40$): 7.1 years (0.7 years) 83% male ASD, 100% Placebo ($n = 40$): 6.3 years (1.2 years) 84% male ASD, 100%	• None reported	
Van der Heijden *et al.*[111] Associated publications: Hoebert *et al.*[112] The Netherlands	Melatonin: fast release, 3 mg (if < 40 kg), 6 mg (if > 40 kg) at 19.00, 4 weeks Control: identical appearing placebo at 19.00, 4 weeks	$N = 107$ Melatonin ($n = 54$): 9.1 years (2.3 years) 66% male ADHD subtype: ADHD-C, 77%; ADHD-I, 17%; ADHD-HI, 4% Placebo ($n = 53$): 9.3 years (1.8 years) 83% male ADHD subtype: ADHD-C, 69%; ADHD-I, 25%; ADHD-HI, 4%	Sleep disturbance • Sleep initiation: sleep onset insomnia Prior interventions • None reported	Unclear

continued

TABLE 32 Study details for pharmacological interventions (*continued*)

Study details and study design	Trial treatments	Participant characteristics: number randomised, mean age (SD); % male, % diagnosis	Type of sleep disturbance/prior interventions	Risk of bias (low, unclear or high)
Crossover trials				
Camfield *et al.*[113] Individualised '*N* of 1 crossover trial' Canada	Melatonin: capsules 0.5 mg for three cases and 1.0 mg for three cases, taken at 18.00, given for 1 week Placebo: capsules, identical-looking to intervention, given for 1 week Ten-week trial in which, for each of the five 2-week intervals, the child was randomised to receive daily placebo or melatonin for the first week with the alternate agent given in the second week	*N* = 6 7.3 years, range 3–13 years 67% male Congenital optic nerve hypoplasia: 17%; moderate spastic quadriplegia 17%; schizencephaly, spastic diplegia and severe learning disabilities, 17%; moderate learning disabilities and severe athetoid cerebral palsy, 17%; developmental disorder with extreme hyperactivity and moderate to severe learning disabilities, 17%; autosomal recessive disorder with cerebellar hypoplasia and tapetoretinal degeneration but with stable moderate to severe learning disabilities, 17%	Sleep disturbance • Sleep initiation: delayed sleep phase • Sleep maintenance: disrupted and fragmented sleep Prior interventions • All parents reported attempting behavioural approaches to nocturnal awakenings and at least one hypnotic agent, without success	High
Dodge and Wilson[114] Associated publications: Hoebert *et al.*[112] USA	Melatonin: capsules, dose 5 mg, taken at 20.00 for 2 weeks Placebo: capsule and filler packaged to be identical to the melatonin capsules, dose 5 mg, taken at 20.00 for 2 weeks Washout period of 1 week	*N* = 36 89 months, range 13 months to 15 years Gender: not reported Cerebral palsy, 75%; autism, 10%; genetic syndrome, 10%; learning disabilities, 5%	Sleep disturbance • 'Chronic sleep problems' Prior interventions • Individuals were excluded if behavioural interventions had not been 'adequately tried'	Unclear

Study details and study design	Trial treatments	Participant characteristics: number randomised, mean age (SD); % male, % diagnosis	Type of sleep disturbance/prior interventions	Risk of bias (low, unclear or high)
Garstang and Wallis[69] UK	Melatonin: dose 5 mg, for 4 weeks (type of dose not reported) Placebo: capsule, dose not reported, for 4 weeks Washout period of 1 week	*N* = 11 8.6 years (3.1 years), range 5–15 years 64% male All ASD plus: mild LD, 27%; moderate LD, 18%; severe LD, 9%; dyspraxia, 18%; no other diagnoses, 27%	Sleep disturbance • Sleep initiation: sleep latency of at least 1 hour after bedtime • Sleep maintenance: night awakenings requiring parental attention, present for 4 nights/week or more in previous 6 months Prior interventions • There had to be a failure of behavioural management techniques to be included. All parents received a standard advice leaflet about establishing good sleep hygiene measures	High
Jain *et al.*[115] Associated publications: Jain *et al.*[116] USA	Melatonin: sustained release, tablet form, dose 9 mg, taken 30 minutes before bedtime for 4 weeks Placebo: tablet form, the same appearance as melatonin tablets, dose not reported Washout period of 1 week	*N* = 11 8.4 years (1.3 years) 70% male Epilepsy type: focal, 70%; generalised, 20%; undetermined, 10%	Sleep disturbance • A score of ≥ 30 on Sleep Behaviour Questionnaire subscales: sleep fragmentation, parasomnia and daytime drowsiness Prior interventions • None reported	High

continued

TABLE 32 Study details for pharmacological interventions (*continued*)

Study details and study design	Trial treatments	Participant characteristics: number randomised, mean age (SD); % male, % diagnosis	Type of sleep disturbance/prior interventions	Risk of bias (low, unclear or high)
Wasdell *et al.*[117] Associated publications: Carr *et al.*[118] Canada	Melatonin: controlled release, capsules, dose 5 mg, taken 20–30 minutes before bedtime for 10 days Placebo: identical capsules to melatonin, dose 5 mg, 20–30 minutes before bedtime for 10 days 'Placebo washout' period of 3–5 days	*N* = 51 7.4 years, range 2.1–17.8 years; 62% Severe intellectual loss, 64%; cerebral palsy, 52%; epilepsy, 46%; visual impairment, 40%; lack of mobility, 36%; ASD, 32%	Sleep disturbance • Sleep initiation: chronic delayed sleep syndrome • Sleep maintenance: impaired sleep maintenance Prior interventions • Prior to randomisation, a paediatric nurse supervised caregivers, while participants received sleep hygiene tailored to their age, development and individual disabilities. Only children whose sleep problems persisted were randomised	Unclear
Weiss *et al.*[70] Canada	Melatonin: short acting, dose 5 mg, taken 20 minutes before bedtime for 10 days Placebo: dose X mg for 10 days The 10-day treatment phases were separated by a 'placebo washout' period of 5 days	*N* = 23 10.29 years, range 6.5–14.7 years 91% male All ADHD. Subtype: inattentive, 4%; ADHD-C, 96%	Sleep disturbance • Sleep initiation: initial insomnia of > 60 minutes Prior interventions • At the start of the study children received a sleep hygiene intervention. Only children who continued to have initial insomnia for > 60 minutes were randomised	Unclear
Wirojanan *et al.*[119] USA	Melatonin: dose 3 mg, taken 30 minutes prior to bedtime for 2 weeks Placebo: dose 3 mg, taken 30 minutes before bedtime for 2 weeks No washout period	*N* = 18 5.5 years (3.6 years), range 2–15.3 years 92% male Fragile X syndrome + ASD, 25%; fragile X syndrome, 25%; ASD, 42%; fragile X syndrome permutation, 8%	Sleep disturbance • 'Sleep problem' Prior interventions • None reported	Unclear

Study details and study design	Trial treatments	Participant characteristics: number randomised, mean age (SD); % male, % diagnosis	Type of sleep disturbance/prior interventions	Risk of bias (low, unclear or high)
Wright *et al.*[106] UK	Melatonin: standard release melatonin, dose 2 mg, taken 30–40 minutes before bedtime. Dose increased by 2 mg every 3 nights to a maximum of 10 mg. Taken for 3 months Placebo: capsules identical to melatonin, dose 2 mg, taken 30–40 minutes before bedtime. Dose increased by 2 mg every three nights to a maximum of 10 mg. Taken for 3 months Washout period of 1 month	*N* = 20 9.0 years (2.9 years), range 4–16 years 80% male Autism, 70%; atypical autism, 20%; Asperger syndrome, 10%	Sleep disturbance • Sleep initiation: sleep latency • Sleep maintenance: excessive night waking or reduced TST Prior interventions • Children were only invited to participate if behaviour management with parenting support had been provided by an experienced clinician and was not successful. Previous interventions were discussed to ensure all participants had been offered adequate behaviour management and parenting support. If participants had not received behaviour management and support, they were referred to the learning disability Child and Adolescent Mental Health Services team. Families that had received appropriate support received a refresher session. Sleep diaries were completed for 1 month and only those who had persistent sleep problems were randomised	Unclear
Melatonin vs. melatonin *Crossover trials*				
Hancock *et al.*[120] UK	Melatonin: 1 × 5 mg plus 1 × 5 mg placebo, 30 minutes before bedtime for 2 weeks Melatonin: 2 × 5 mg of melatonin (10 mg in total), taken 30 minutes before bedtime for 2 weeks Washout period of 2 weeks	*N* = 8 12.1 years (10.0 years), range 1.5–31 years 57% male All tuberous sclerosis Of participants aged < 19 years 6.9 years (4 years), range 1.5–11 years; 40% All tuberous sclerosis	Sleep disturbance • Quine sleep index score of at least 6 (of a possible 8) Prior interventions • None reported	High

continued

TABLE 32 Study details for pharmacological interventions (*continued*)

Study details and study design	Trial treatments	Participant characteristics: number randomised, mean age (SD); % male, % diagnosis	Type of sleep disturbance/prior interventions	Risk of bias (low, unclear or high)
Jan *et al.*[121] Canada	Melatonin: sustained-release melatonin, variable doses from 2–10 mg, taken 30 minutes before bedtime, 11 days Control: fast-release melatonin, variable does from 2–10 mg, taken for 11 days No washout period	*N* = 16 Age range: 4–21 years *N* = 15 eligible participants (under 18 years) 9.3 years, range 4–16 years Gender: not reported Multidisabled but not reported separately for RCT	Sleep disturbance • 'Chronic sleep–wake disorders' Prior interventions • None reported	High

ADHD-C, attention deficit hyperactivity disorder – combined type; ADHD-I, attention deficit hyperactivity disorder – inattentive type; ADHD-HI, attention deficit hyperactivity disorder – hyperactive impulsive type; CBT, cognitive–behavioural therapy; DD, developmental delay; LD, learning disability.

Appendix 6 Study quality: studies evaluating melatonin

TABLE 33 Summary of quality assessment using the Cochrane risk of bias for RCTs tool[62,a]

Study	Domain						
	1: adequate sequence generation?	2: allocation concealment?	3: blinding?	4: incomplete outcome data addressed?	5: free of selective reporting?	6: free of other bias?	Additional questions for crossover trials
Melatonin vs. placebo							
Parallel trials							
Appleton *et al.*[48]	Yes Computer-generated sequence	Yes Pharmacy led	Yes Double blinding, using identical capsules and central allocation. In two cases participants were unblinded in order to treat serious adverse events	Yes Similar numbers lost in each arm for similar reasons. Data analysed on intention-to-treat basis	Yes Protocol available, amendments detailed, all outcomes reported	Yes No other bias obvious	N/A
Cortesi *et al.*[110]	Yes Computer-generated sequence	Unclear Allocation was done by someone independent of treatment personnel and via computer, but it is not clear if the allocation was concealed to the independent person (e.g. via opaque envelopes)	Unclear Authors argue that researchers and participants did not know which arm they were in (there were four arms in total). Although this is plausible for the melatonin only and placebo-only arms, in which the capsules were identical, it is less plausible for the melatonin combined with CBT and the CBT only, as they would obviously know if they were receiving CBT. Similarly, the principal investigators (PIs) observed tapes of the CBT for adherence to protocol, so they would know at least who was receiving CBT (even if they could not know if they were receiving this alone or with melatonin), so this is in agreement with the above judgement	No None were lost to follow-up but 16 dropped out owing to difficulties in administering medication, non-compliance and lack of improvement. An additional 10 were excluded from analysis owing to missing actigraphy data. The final analysis excluded all above. There were no details of the potential implications of missing data/dropouts. All six dropouts in placebo group were because of lack of improvement; none of the dropouts in the other groups were for this reason	Unclear No protocol is referenced in the paper and one was not found on searching. The study reports outcomes that one would expect. However, table 3 reports two outcomes that are not referred to anywhere else in the paper (Naptime and Bedtime). It is not clear what they are In the methods section, the paper only states what the primary outcomes were and so it is presumed that Naptime and Bedtime are secondary, but this is not stated so there is a low degree of selective reporting, as the authors state that groups and clinicians were unaware of what they were receiving and both groups were treated as per an identical protocol	Yes	N/A

Study	Domain						
	1: adequate sequence generation?	2: allocation concealment?	3: blinding?	4: incomplete outcome data addressed?	5: free of selective reporting?	6: free of other bias?	Additional questions for crossover trials
Van der Heijden *et al.*[111]	Unclear Randomisation in blocks of four to keep number in each treatment group closely balanced. No details of how sequence generated	Yes Central allocation. Performed by a hospital pharmacist who was not connected to study	Yes Reported as double-blind and the placebo tablets looked identical to intervention tablets. Investigators and participants unware of allocation. Code broken after all treatment completed and data recorded	Yes No loss to follow-up. One child in placebo group and one in the melatonin group were withdrawn prior to treatment because they started other treatment without permission	Unclear Protocol available on the International Standard Randomised Controlled Trial Number website (although it says it was retrospectively registered). Outcomes listed on trial record are: (primary) sleep onset, latency, total sleep duration (acting and sleep log) and Dim Light Melatonin Onset and (secondary) sustained attention and response inhibition, severity of ADHD symptoms, quality of life and side effects. The study paper reports these outcomes but also reports that difficulty falling asleep (rated in severity) was a primary outcome (p. 235). CBCL (behavioural problems) was also reported (which may be the measure of ADHD severity listed in protocol but this is not clear) An additional file that includes all raw data for study outcomes is available	Yes The number of missing data on behaviour and quality-of-life measures was large but authors suggest that the risk of bias in the analysis is small because the missing data are equal in both groups	N/A
Crossover trials							
Camfield *et al.*[113]	Unclear No details are provided in the paper	Unclear No details are provided in the paper	Unclear Participants were blinded, although not clear how. Not clear if researchers blinded at analysis	Yes No loss to follow-up and data presented for all six participants	Unclear No protocol is referenced in the paper and one was not found on searching. Three outcome measures are used: average hours of sleep per 24 hours, number of awakenings between 9 p.m. and 7 p.m. per day and number of nights with no arousals from 8 p.m. to 7 a.m. The reported 'nights without awakening' was 10 p.m. to 7 a.m. rather than 9 p.m. to 7a.m. Not clear whether this was a typo or the outcome measure changed	No Small sample and only descriptive data are reported	

continued

TABLE 33 Summary of quality assessment using the Cochrane risk of bias for RCTs tool[62,a] (*continued*)

Study	Domain						
	1: adequate sequence generation?	2: allocation concealment?	3: blinding?	4: incomplete outcome data addressed?	5: free of selective reporting?	6: free of other bias?	Additional questions for crossover trials
Dodge and Wilson[114]	Unclear Randomisation performed by pharmacy personal but sequence generation not described	Unclear Randomisation and packaging of capsules done by research pharmacy personal but it is not clear if these research pharmacy personal were part of research team, and no detail is provided about whether or not allocation was concealed	Yes Identical packaging. Parents asked to guess which treatment their children were on prior to unblinding. Randomisation by research pharmacists, not study team	Unclear No flow chart. In total, 20 out of 36 participants who enrolled completed the study but it was not clear how missing data were addressed. Losses to follow-up were reported as having no significant difference on age, primary diagnosis, epilepsy or vision impairment compared with those completing study. Non-completion was because of lost sleep logs ($n = 1$), lost medication ($n = 2$), changed mind about participating ($n = 4$), intercurrent illness ($n = 2$), family emergency ($n = 1$), family lost to follow-up ($n = 3$) and medication not working ($n = 2$)	Unclear No protocol was referenced in the paper and one was not found after searching. The outcomes reported are those that are expected. However, without protocol, it is difficult to be certain	Unclear Details of sample characteristics between treatment and placebo at baseline are not reported, making the comparability unclear	N/A
Garstang and Wallis[69]	Yes Random numbers table	Yes Pharmacy led	Unclear Reported as double blind but there were few details as to how this was achieved. There was no reference to treatment and placebo capsules being identical	No Dropouts: after drugs recall following empty placebo capsules, $n = 3$; owing to a house move, $n = 1$; and on the start of a child protection enquiry, $n = 1$. No detail on whether last two were receiving placebo or melatonin. Data were analysed for trial completers only	Unclear No protocol was referenced in the paper and one was not found after searching. The outcomes reported are those that are expected. However, without a protocol, it is difficult to be certain	No There was an assumption that the child was asleep (and logged as asleep) if parents were not disturbed. Child could have been awake but did not disturb the parent. There was also a drugs recall and the trial was stopped early, so it is not clear if and how this affected subsequent procedure and blinding of treatment	Was use of a crossover design appropriate? Yes Is it clear that the order of receiving treatments was randomised? Yes Can it be assumed that the trial was not biased from carry-over effects? Washout period used, but no analysis of carry-over effect Are unbiased data available? There does not appear to be any analysis of difference between the treatment and control groups

Study	Domain						
	1: adequate sequence generation?	2: allocation concealment?	3: blinding?	4: incomplete outcome data addressed?	5: free of selective reporting?	6: free of other bias?	Additional questions for crossover trials
Jain *et al.*[115]	Yes Computer-randomised number generator	Yes Pharmacy led	No The appearance of capsules was identical but the statistician and pharmacist were unblinded	Unclear In total, 1 out of 11 did not complete the study owing to being unable to swallow the capsules The limitations section states that there were some secondary outcome data lost for two participants and that this may have had an impact on the results. It is not clear what the missing data or impacts were	No Protocol is available on clinical trials register. Outcomes listed on register include: (primary) sleep efficiency and improved lapse time on psychomotor vigilance task and (secondary) improvement in eleptiform discharges on electroencephalography and seizure frequency. However, in the paper, more outcomes are reported. The primary outcomes in the paper are SOL and wakefulness. Twenty secondary outcomes are listed, including the one listed as primary (sleep efficiency) on the trials register	Yes	Was use of a crossover design appropriate? Yes Is it clear that the order of receiving treatments was randomised? Yes Can it be assumed that the trial was not biased from carry-over effects? Carry-over effects observed for two REM variables and so adjusted analysis Are unbiased data available? When carry-over effects are not observed, authors report using Wilcoxon rank-sum test and two sample tests

continued

TABLE 33 Summary of quality assessment using the Cochrane risk of bias for RCTs tool[62,a] (*continued*)

Study	Domain						
	1: adequate sequence generation?	2: allocation concealment?	3: blinding?	4: incomplete outcome data addressed?	5: free of selective reporting?	6: free of other bias?	Additional questions for crossover trials
Wasdell *et al.*[117]	Unclear Blocked randomisation method led by pharmacy, 'in which every four patients had equal probability of receiving either of the two treatment sequences' (p. 58). However, no detail given on how the randomisation sequence was generated	Yes Pharmacy led	Yes Patients, caregivers, the study investigator and clinical staff were blinded to the medication randomisation. Unblinding occurred at the end of the study	Yes Overall, 50 out of 51 randomised participants completed trial. One withdrew owing to illness. Only the 50 who completed the trial were included in the analysis	Unclear No protocol was referenced in the paper and one was not found after searching. The outcomes reported are those that are expected. However, without a protocol, it is difficult to be certain	Yes	Was use of a crossover design appropriate? Yes Is it clear that the order of receiving treatments was randomised? Yes Can it be assumed that the trial was not biased from carry-over effects? Washout period used and authors undertook analysis to test for carry-over effect and report that there were none Are unbiased data available? Yes. Paired *t*-test used

Study	Domain						
	1: adequate sequence generation?	2: allocation concealment?	3: blinding?	4: incomplete outcome data addressed?	5: free of selective reporting?	6: free of other bias?	Additional questions for crossover trials
Weiss *et al.*[70]	Unclear No details are provided	Yes Pharmacy led	Unclear Authors state that all participants and study personal blinded to the order of treatment during randomisation phase. This implies that blinding may not have extended after randomisation (e.g. at data collection and analysis). However, no further detail is given so it is difficult to judge this	Yes 3/22 discontinued because of protocol violations but outcomes unchanged whether included or not (were excluded in results presented in paper)	Unclear No protocol referenced in the paper and one was not found after searching. The outcomes reported are those that are expected. However, without protocol, it is difficult to be certain. Furthermore, CADS-P was not reported, other than that no significant change was observed; therefore, the data cannot be included in a meta-analysis	Yes	Was use of a crossover design appropriate? Yes Is it clear that the order of receiving treatments was randomised? Not clear Can it be assumed that the trial was not biased from carry-over effects? Washout period used and authors undertook analysis to test for carry-over effect and report there were none Are unbiased data available? Yes. Paired analysis used

continued

TABLE 33 Summary of quality assessment using the Cochrane risk of bias for RCTs tool[62,a] (*continued*)

Study	Domain						
	1: adequate sequence generation?	2: allocation concealment?	3: blinding?	4: incomplete outcome data addressed?	5: free of selective reporting?	6: free of other bias?	Additional questions for crossover trials
Wirojanan *et al.*[119]	Unclear No details are provided	Unclear Authors say that the allocation was concealed but do not say how	Yes Authors say that assignment to the treatment condition was concealed until the end of the study. Allocation key with investigator (locked in file) until the end of the study	Yes In total, 12 out of 18 participants completed trial. No details of characteristics of the six who did not complete. Reasons were sleep diaries not completed/actigraphy data not readable ($n = 2$ placebo, $n = 1$ melatonin); actigraphy watch not worn during first treatment phase (melatonin) ($n = 1$); actigraphy watch taken off in first phase (placebo) ($n = 1$); study protocol not followed ($n = 1$). Data only from the 12 were analysed Missing output data dealt with by complete-case analysis and last observation carried forward	Unclear No protocol was referenced in the paper and one was not found after searching. The outcomes reported are those that are expected. However, without a protocol, it is difficult to be certain	Unclear Authors report that normality assumption for parametric paired test was violated owing to highly skewed data and presence of outliers. The authors present both parametric and non-parametric analyses. The authors imply in the discussion that some families may have been practising sleep hygiene, with variability in this. There is no further information about this but the implication is that this was possible and not minimised or controlled. Without any further information (e.g. how many families practised sleep hygiene), it is difficult to judge whether or not this has introduced bias	Was use of a crossover design appropriate? Yes Is it clear that the order of receiving treatments was randomised? No Can it be assumed that the trial was not biased from carry-over effects? No. No washout period was used between crossover. The authors say that this is because of the half-life of melatonin but is this conclusive? Different studies have used different approaches – some having a washout period and others not. It is not clear or certain if this could present a bias Are unbiased data available? Paired *t*-tests used, both parametric and non-parametric

Study	Domain						
	1: adequate sequence generation?	2: allocation concealment?	3: blinding?	4: incomplete outcome data addressed?	5: free of selective reporting?	6: free of other bias?	Additional questions for crossover trials
Wright *et al.*[106]	Unclear No details are provided	Yes Remote pharmacy led	Yes Double-blind randomisation was undertaken by someone with no contact with the research team and the capsules were identical	Yes There were four withdrawals (three in melatonin first, one in placebo first, but two while taking melatonin and two while taking placebo). Reasons: too difficult for parents to administer medication, $n = 1$; parents unable to complete sleep diary questionnaire measures, $n = 1$; very significant sleep benefits early in first arm, $n = 1$; and apparent ineffectiveness of medication, $n = 1$. The last completed one arm of trial and so data were included in some of analyses. This participant was in melatonin arm first and another clinician prescribed another drug Only one withdrawal (early very significant effects) was likely to be related to the true outcome	Unclear No protocol was referenced in the paper and one was not found after searching. The outcomes reported are those that are expected. However, without a protocol, it is difficult to be certain	Yes	Was use of a crossover design appropriate? Yes Is it clear that the order of receiving treatments was randomised? No Can it be assumed that the trial was not biased from carry-over effects? A washout period of 1 month used Are unbiased data available? Paired *t*-tests were used
Melatonin vs. melatonin *Crossover trials*							
Hancock *et al.*[120]	Unclear The hospital pharmacy led randomisation but it is not clear how the sequence was generated The pharmacy department generated random numbers that determined whether individuals started with 5 mg or 10 mg of melatonin. This is assessed as LOW risk/Yes	Yes Pharmacy led	Yes The study was double blind, identical capsules were used and unblinding occurred after completion of trial	Unclear In total, one out of eight sets of data was lost in the mail. There are no details of lost patient characteristics and the implications of this There is no detail of how other missing data are dealt with	Unclear No protocol was referenced in the paper and one was not found after searching. The outcomes reported are those that are expected. However, without a protocol, it is difficult to be certain	No It had a small sample, but it is a pilot study	Was use of a crossover design appropriate? Yes Is it clear that the order of receiving treatments was randomised? Yes Can it be assumed that the trial was not biased from carry-over effects? A washout period used but no analysis of carry-over effect Are unbiased data available? Paired *t*-tests used

continued

TABLE 33 Summary of quality assessment using the Cochrane risk of bias for RCTs tool[62,a] (*continued*)

Study	Domain						
	1: adequate sequence generation?	2: allocation concealment?	3: blinding?	4: incomplete outcome data addressed?	5: free of selective reporting?	6: free of other bias?	Additional questions for crossover trials
Jan *et al.*[121]	Unclear No details are provided in the paper	Unclear No details are provided in the paper	Unclear Paper reports that both investigators and caregivers were blinded but no detail is given as to how (e.g. identical packs)	Yes No loss to follow-up	No No protocol was referenced and one was not found after searching. However, the only outcome reported is changes in sleep pattern, which is reported as whether or not there was a response to treatment (with little detail on what exactly was measured). There are no data presented regarding what this response was and it is, thus, rather vague. Perhaps this is because it was a dose-finding RCT, but even so, the lack of detail on the outcome makes the reporting appear selective and lacks transparency	No Participants had already been treated with another form of melatonin. It is not clear how the 16 participants were selected; for example, whether or not they fit eligibility criteria	Was use of a crossover design appropriate? Yes Is it clear that the order of receiving treatments was randomised? Yes Can it be assumed that the trial was not biased from carry-over effects? No, as there was no washout period between crossover. There was no analysis of carry-over effect Are unbiased data available? No. The type of analysis used was not clear

CADS-P, Conners Attention Deficit Scale Parent version; CBT, cognitive–behavioural therapy; N/A, not applicable; REM, rapid eye movement.

a Yes, low risk of bias; unclear, unclear risk of bias; no, high risk of bias.

Appendix 7 Child sleep-related outcomes in trials comparing melatonin with placebo

TABLE 34 Child sleep-related outcomes in trials comparing melatonin with placebo

Study	Child sleep-related outcome assessed	Method of assessment[a]	Definitions[b]
Global measures and composite scores			
Appleton *et al.*[48]	Sleep efficiency	Actigraphy	Number of minutes spent sleeping in bed/total number of minutes spent in bed × 100
	TST	Actigraphy, sleep diary	The amount of time between the time that the child went to sleep and the time that the child woke up the following morning minus any night-time awakenings
Camfield *et al.*[113]	TST	Sleep diary	Average sleep per day
Cortesi *et al.*[110]	CSHQ	CSHQ (total score and subscales)	A 33-item parent questionnaire that includes items relating to a number of key sleep domains, which are grouped into the following subscales: bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night wakings, parasomnias, sleep disordered breathing and daytime sleepiness. The authors provide a reference
	Sleep efficiency	Actigraphy	No definition provided, other than that 'these variables were averaged over 7 nights for each assessment phase'[110]
	TST	Actigraphy[c]	No definition provided, other than that 'these variables were averaged over 7 nights for each assessment phase'[110]
	WASO	Actigraphy[c]	Inclusion criteria defined WASO as wake after sleep onset > 30 minutes that occurred on ≥ 3 nights a week
Van der Heijden *et al.*[111]	Sleep efficiency	Actigraphy[c]	Percentage of time spent asleep in the period from lights out until the time of leaving bed
	TST	Actigraphy[c]	Actual amount of sleep, calculated as the period from sleep onset to wake up time minus estimated time awake in the period from sleep onset until awake time
Dodge and Wilson[114]	TST	Sleep log	Duration of sleep
Garstang and Wallis[69]	TST	Sleep chart	No definition provided
Jain *et al.*[115]	Sleep efficiency	Polysomnography, actigraphy[d]	'The American Academy of Sleep Medicine standard definition was used'[115]
	TST	Polysomnography, actigraphy, sleep diary	'The American Academy of Sleep Medicine standard definition was used'[115]
	WASO	Polysomnography, actigraphy[d]	The sum of wake time in minutes from sleep onset to the final awakening

continued

TABLE 34 Child sleep-related outcomes in trials comparing melatonin with placebo (*continued*)

Study	Child sleep-related outcome assessed	Method of assessment[a]	Definitions[b]
	Sleep Behaviour Questionnaire	Sleep Behaviour Questionnaire	Sensory Behaviour Questionnaire includes a set of six questions related to sleep–wake habits and a 29-item Likert-type rating scale. The subscales include parasomnias, parent/child interaction, sleep fragmentation, daytime drowsiness and bedtime difficulties. The Sensory Behaviour Questionnaire total score is considered a global index of sleep problems, with higher scores representing more sleep problems
Wasdell *et al.*[117]	Sleep efficiency	Actigraphy, somnolog	No definition provided
	TST	Actigraphy, somnolog	For somnolog-measured TST: 'Total night-time sleep as recorded on care-giver completed somnologs which provided a running record of the time when the child was asleep or awake during the relevant measurement period'[117]
	Longest sleep episode	Somnolog, actigraphy	No definition provided
Weiss *et al.*[70]	TST	Actigraphy, somnolog	Total sleep duration
Wirojanan *et al.*[119]	TST	Actigraphy[c]	The time from sleep onset to wake-up time minus the time awake during the night
Wright *et al.*[106]	TST	Sleep diary	No definition provided
Sleep initiation			
Appleton *et al.*[48]	SOL	Actigraphy, sleep diary	The time taken to fall asleep; the number of minutes between lights out/'snuggle down' time and sleep start time
Cortesi *et al.*[110]	SOL	Actigraphy[c]	Inclusion criteria defined SOL as mixed sleep onset and maintenance insomnia
	Bedtime	Actigraphy[c]	No definition provided
Van der Heijden *et al.*[111]	Difficulty falling asleep	Sleep log	Averaged score (over seven days) on an item asking parents how difficult it was for the child to fall asleep in the evening (1 = not difficult; 5 = very difficult)
	Sleep onset	Actigraphy[c]	Start of a period of at least 10 minutes of consecutively recorded immobile actigraphy data following lights out
	SOL	Actigraphy[c]	Time from lights out until sleep onset
Dodge and Wilson[114]	SOL	Sleep log	Time taken to fall asleep
Garstang and Wallis[69]	SOL	Sleep chart	No definition provided
Jain *et al.*[115]	SOL	Polysomnography, actigraphy (data not reported)	The time in minutes from lights out to sleep onset
	Bedtime	Sleep diary	No definition provided
Wasdell *et al.*[117]	SOL	Actigraphy, somnolog	No definition provided
Weiss *et al.*[70]	SOL	Actigraphy, somnolog	The amount of time between when the child was put to bed and when he or she fell asleep
Wirojanan *et al.*[119]	SOL	Actigraphy[c]	The time from bedtime to sleep onset time
	Sleep onset	Actigraphy[c]	The clock time that the child fell asleep
Wright *et al.*[106]	SOL	Sleep diary	Time from start of bedtime routine to sleep
	Sleep latency	Sleep diary	Time from drug to sleep

TABLE 34 Child sleep-related outcomes in trials comparing melatonin with placebo (*continued*)

Study	Child sleep-related outcome assessed	Method of assessment[a]	Definitions[b]
Sleep maintenance			
Camfield *et al.*[113]	Number of night wakings	Sleep diary	Total number of awakenings between 9.00 p.m. and 7.00 a.m. per day/the number of days of complete data
	Nights without awakening	Sleep diary	Nights without awakening between 10.00 p.m. and 7.00 a.m. per day/the number of days of complete data
Van der Heijden *et al.*[111]	Wake time	Actigraphy	Last epoch of actigraphically assessed immobility before the start of a 10-minute consecutive period of activity around the time of leaving bed
	Non-specified night-time sleep disturbance: moving time	Actigraphy	Percentage of time spent moving during the assumed sleep period
Dodge and Wilson[114]	Number of night wakings	Sleep log	No definition provided
Garstang and Wallis[69]	Number of night wakings	Sleep chart	No definition provided
Jain *et al.*[115]	Wake time	Sleep diary	No definition provided
Wasdell *et al.*[117]	Number of night wakings	Actigraphy, somnolog	No definition provided
Wirojanan *et al.*[119]	Number of night wakings	Actigraphy[c]	No definition provided
Wright *et al.*[106]	Number of night wakings	Sleep diary	No definition provided
Sleep scheduling			
Cortesi *et al.*[110]	Naptime	Actigraphy[c]	No definition provided
Other outcomes			
Van der Heijden *et al.*[111]	Interdaily stability	Actigraphy	Degree of resemblance between the activity patterns on individual days (range from 0 to 1, higher values indicating more stable rhythms)
	Interdaily variability	Actigraphy	Fragmentation of periods of rest (or sleep) and activity (or wakefulness) (range from 0 to 2, with higher values indicating more fragmented rhythms)
	L5 (average activity during the least active 5 hours)	Actigraphy	Average activity during the least active 5-hour period in the average 24-hour activity rhythm
Jain *et al.*[115]	Arousal	Arousal index	American Academy of Sleep Medicine standard definitions were used for arousal index. The authors provide a reference
	Percentage of sleep stages	Sleep diary	American Academy of Sleep Medicine standard definitions were used for percentage of sleep stages (N1, N2, N3 or REM %). The authors provide a reference

REM, rapid eye movement.
a Various terms used for sleep diaries (logs, charts, somnologs).
b As defined by study author.
c Only reports actigraphy data; however, a sleep diary/log was used to provide verification.
d Only reports polysomnography data.

Appendix 8 Child sleep-related outcomes in trials comparing melatonin with melatonin

TABLE 35 Child sleep-related outcomes in trials comparing melatonin with melatonin

Study	Child sleep-related outcome assessed	Method of assessment[a]	Definitions[b]
Global measures and composite scores			
Hancock *et al.*[120]	TST	Sleep diary	No definition provided
Jan *et al.*[121]	Changes in sleep pattern	Sleep charts and parental history	Based on sleep onset, the number of awakenings, duration of sleep and how the children behaved the following day
Sleep initiation			
Hancock *et al.*[120]	Bedtime settling: SOL	Sleep diary	Time taken to fall asleep
Sleep maintenance			
Hancock *et al.*[120]	Night waking: number of night wakings	Sleep diary	The mean number of awakenings each night

a Various terms used for sleep diaries (logs, charts, somnologs).
b As defined by study author.

Appendix 9 Study details for non-pharmacological interventions

TABLE 36 Study details for non-pharmacological interventions

Study details and study design	Intervention details	Participant characteristics: number randomised (*N*), mean age (SD); % male; % diagnosis	Type of sleep disturbance/prior sleep interventions	Risk of bias (low, unclear or high)
Parent-directed comprehensive tailored interventions *Parallel trials*				
Beresford *et al.*[21] Associated publications: Beresford *et al.*[136] UK	Intervention: two face-to-face sessions for the assessment and development of a sleep management strategy and parent training were given. Parents were supported to implement the strategy via telephone calls Control: the usual approach. As for the intervention but implementation support was delivered via home visits	*N* = 13 Home visits (*n* = 6): • 2.67 years (0.82 years) • 100% male • ASC, 50%; LD, 16.7%; physical or sensory disability, 16.7%; unknown/ awaiting diagnosis, 16.7% Telephone support (*n* = 7): • 2.86 years (1.07 years) • 71% male • ASC, 71%; LD, 14%; and physical or sensory disability, 14%	Sleep disturbance • Sleep initiation: bedtime resistance, sleep anxiety • Sleep maintenance: night wakings Prior interventions • Not reported	High
Hiscock *et al.*[138] Associated publications: Papadopoulos *et al.*[139] Australia	Intervention: one session was given for the assessment and development of a sleep management strategy and parent training. Parents were supported to implement the strategy via one face-to-face session and one telephone call Control: usual care	*N* = 244 Behavioural programme (*n* = 122): • 10.3 years (1.8 years) • 84% male • ADHD, 100%; LD, 34%; autism spectrum or Asperger syndrome, 23% Usual care (*n* = 122): • 9.9 years (2.1 years) • 86% male • ADHD, 100%; LD, 34%; and autism spectrum or Asperger syndrome, 27%	Sleep disturbance • 'Moderate/severe sleep problems' including: ○ Sleep initiation: sleep onset association disorder, limit setting disorder, delayed sleep phase, idiopathic or psychophysiological insomnia Prior interventions • None reported: an exclusion criterion was if children were already receiving specialist sleep input	High

Study details and study design	Intervention details	Participant characteristics: number randomised (*N*), mean age (SD); % male; % diagnosis	Type of sleep disturbance/prior sleep interventions	Risk of bias (low, unclear or high)
Johnson *et al.*[107] Associated publications: Turner[140] USA	Intervention: one session for assessment and the development of sleep management strategy was given; there were five sessions for training the parent in the strategy. Implementation support was delivered via one face-to-face session Control: non-sleep-related parent education delivered in an identical manner to the intervention group	*N* = 40 Behavioural parent training programme (*n* = 20, data on *n* = 15): • 3.51 years (0.98 years) • 73% male • Autism, 80%; and ASD, 20% Non-sleep-related parent education (*n* = 20, data on *n* = 18): • 3.6 years (1.12 years) • 83% male • Autism, 94%; and ASD, 6%	Sleep disturbance • Sleep initiation: bedtime resistance, delayed sleep onset and sleep association problems • Sleep maintenance: night-time wakings, morning waking before 5:00 a.m. and when the child disturbed the parent or entered the parents' bedroom Prior interventions • Not reported, but exclusion criteria included children using sleep supplements	High
Moss *et al.*[124] Associated publications: O'Connell *et al.*[141,142] Australia	Intervention: two parent training workshops, followed by a home visit for the assessment and development of a sleep management strategy were given. Implementation support was delivered via one home visit followed by telephone calls as needed Control: waiting list control	*N* = 26 Intervention and control not reported separately • Overall 11.74 years (2.53 years) • Gender: not reported • ASD, 58%; Angelman, Down and other syndromes, 19%; intellectual disability and global developmental delay, 11%; blindness, 4%; and unspecified, 8%	Sleep disturbance • Sleep initiation: settling • Sleep maintenance: problems with night waking, sleep duration/ regularity, co-sleeping and early waking • Sleep scheduling: excessing daytime sleepiness • Other: snoring Prior interventions • One child taking melatonin, although it is not clear if this is for sleep	High

continued

TABLE 36 Study details for non-pharmacological interventions (*continued*)

Study details and study design	Intervention details	Participant characteristics: number randomised (*N*), mean age (SD); % male; % diagnosis	Type of sleep disturbance/prior sleep interventions	Risk of bias (low, unclear or high)
Sciberras *et al.*[125] Associated publications: Sciberras *et al.*,[143] Fulton *et al.*[144] and Sciberras and Rinehart[145] Australia	Intervention: two sessions for the assessment and development of a sleep management strategy and training parents in the strategy were given. Implementation support was delivered via a single telephone call followed by further face-to-face session if needed Control: a single session for the assessment and development of a sleep management strategy and training parents in the strategy was given. No implementation support was given	*N* = 27 Extended behavioural programme (*n* = 14): • 12.1 years (2.2 years) • 71% male • ADHD, 100% Brief behavioural programme (*n* = 13): • 10.9 years (2.5 years) • 77% male • ADHD, 100%	Sleep disturbance • 'Moderate/severe sleep problem' including: ○ Sleep initiation – delayed sleep phase, limit setting disorder, anxiety, sleep onset association disorder, insomnia Prior interventions • Not reported	High
Before-and-after studies				
Austin *et al.*[123] Australia	Two parent training workshops were given, followed by a home visit for the assessment and development of sleep management strategy, followed by a third workshop. Implementation support was delivered via one face-to-face session and a telephone call	*N* = 8 3.9 years 100% male Pervasive developmental disorder: • 38% male • ASD, 50%; and unknown, 12%	Sleep disturbance • Sleep initiation: bedtime resistance. Duration of sleep problems ranged from 4 months to 2 years • Sleep maintenance: night waking, co-sleeping, early waking, irregular sleep–wake cycle Prior interventions • Not reported	High

Study details and study design	Intervention details	Participant characteristics: number randomised (*N*), mean age (SD); % male; % diagnosis	Type of sleep disturbance/prior sleep interventions	Risk of bias (low, unclear or high)
Beresford *et al.*[21] Associated publications: Beresford *et al.*[136] and Stuttard *et al.*[45,137] UK	Two sessions were given for the assessment and development of sleep management strategy and training the parents in the strategy. Implementation support was delivered via fortnightly face-to-face sessions	*N* = 12 2.88 years (1.25 years) 50% male ASC, 25%; LD, 25%; and unknown/ awaiting diagnosis, 50%	Sleep disturbance • Sleep initiation: bedtime resistance and sleep anxiety • Sleep maintenance: night wakings Prior interventions • Not reported	High
Quine and Wade[146] Associated publications: Wade and Wade[147] UK	Two sessions were given for the assessment and development of a sleep management strategy and training parents in the strategy. Implementation support delivered via face-to-face sessions	*N* = 25 Range 3–21 years 69% male LD, 100%	Sleep disturbance • Sleep initiation: night settling problems usually ≥ 3 times a week • Sleep maintenance: night waking usually ≥ 3 times a week, limited hours sleep usually ≥ 3 times a week Prior interventions • Not reported	High
Weiskop *et al.*[126] Before-and-after study with multiple baseline Australia	Four sessions were given for the assessment and development of a sleep management strategy and training parents in the strategy. Implementation support (via telephone calls) was delivered from outset of intervention. It continued after training sessions completed with face-to-face session and further telephone calls	*N* = 13 • 5.1 years, range 1–9 years • 77% male • ASD: 46% (autism, 38%; Asperger syndrome, 8%; and fragile X syndrome, 54%)	Sleep disturbance • Sleep initiation: difficulty settling and bed refusal • Sleep maintenance: night waking, co-sleeping or waking early Prior interventions • One child was taking medication for behaviour and sleep problems	High

continued

TABLE 36 Study details for non-pharmacological interventions (*continued*)

Study details and study design	Intervention details	Participant characteristics: number randomised (*N*), mean age (SD); % male; % diagnosis	Type of sleep disturbance/prior sleep interventions	Risk of bias (low, unclear or high)
Parent-directed comprehensive non-tailored interventions *Parallel trials*				
Adkins *et al.*[127] Associated publications: Malow *et al.*[148] USA	Intervention: training curriculum contained in a booklet provided to parents Control: no booklet provided	*N* = 36 Overall age 6.4 years (2.6 years) Sleep education pamphlet (*n* = 18): • 2–5 years, 50%; 6–10 years, 50% • 56% male • Autism, 89%; Asperger syndrome, 11%; and PDD-NOS, 0% Control (no pamphlet) (*n* = 18): • Aged 2–5 years, 33%; aged 6–10 years, 67% • 78% male • Autism, 72%; Asperger syndrome, 22%; and PDD-NOS, 6%	Sleep disturbance • Sleep initiation: SOL of at least 30 minutes for ≥ 3 nights/week Prior interventions • Not reported	High
Montgomery *et al.*[49] Associated publications: Montgomery *et al.*[49] UK	Intervention a: a training curriculum was contained in a booklet provided to parent Intervention b: a training curriculum (content identical to intervention a) was delivered via a face-to-face session Control: there was no intervention for 6 weeks, at which point they were re-randomised into an active treatment group	*N* = 66 Overall age range: 27–101 months 64% male Brief treatment (*n* = 22): • Age: not reported • Gender: not reported • (Not mutually exclusive): autism, 18%; Down syndrome, 9%; global developmental delay, 5%; epilepsy, 5%; other, 20%; no diagnosis, 46%	Sleep disturbance Present for at least 3 months and not because of physical problem: • Sleep initiation: settling problems ≥ 3 times a week in which the child takes more than 1 hour to settle and is disturbing the parents • Sleep maintenance: night waking ≥ 3 times a week and when the child disturbs parents and/or goes into parents' room/bed	High

Study details and study design	Intervention details	Participant characteristics: number randomised (*N*), mean age (SD); % male; % diagnosis	Type of sleep disturbance/prior sleep interventions	Risk of bias (low, unclear or high)
		Conventional treatment ($n = 34$): • Age: not reported • Gender: not reported • (Not mutually exclusive): autism, 35%; Down syndrome, 15%; global developmental delay, 10%; epilepsy, 5%; other, 20%; no diagnosis, 20% Control ($n = 26$): • Age: not reported • Gender: not reported • (Not mutually exclusive): autism, 42%; Down syndrome, 8%; global developmental delay, 8%; epilepsy, 8%; other, 15%; no diagnosis, 17%	Prior interventions • Not reported	
Malow *et al.*[128] USA	Intervention: the training curriculum was delivered via two group-delivered sessions. Implementation support was delivered via telephone calls Control: training curriculum delivered via single, face-to-face session. Implementation support was delivered via telephone calls	$N = 80$ Group-delivered parent education programme ($n = 39$): • 5.9 years (2.8 years) • 64% male • Autism, 67%; Asperger syndrome, 13%; and PDD-NOS, 5% Individual-delivered parent education programme ($n = 41$): • 5.6 years (2.6 years) • 95% male • Autism, 78%; Asperger syndrome, 27%; and PDD-NOS, 9%	Sleep disturbance • Sleep initiation: SOL > 30 minutes for ≥ 3 nights/week Prior interventions • Not reported	High

continued

TABLE 36 Study details for non-pharmacological interventions (*continued*)

Study details and study design	Intervention details	Participant characteristics: number randomised (*N*), mean age (SD); % male; % diagnosis	Type of sleep disturbance/prior sleep interventions	Risk of bias (low, unclear or high)
Before-and-after studies				
Beresford *et al.*[21] Associated publications: Beresford *et al.*[136] and Stuttard *et al.*[45,137] UK	Group delivery of a training curriculum over four sessions	*N* = 22 8.91 years (3.25 years) 50% male ASC, 64%; LD, 27%; physical or sensory disability, 4%; and no diagnosis, 4%	Sleep disturbance • Sleep initiation: bedtime resistance, sleep anxiety and night wakings Prior interventions • Not reported	High
Beresford *et al.*[21] Associated publications: Beresford *et al.*[136] and Stuttard *et al.*[45,137] UK	A training curriculum delivered via a single half-day workshop	*N* = 25 7 years (3.30 years) 64% male ASC, 36%; LD, 16%; physical or sensory disability, 16%; and ASC other, 20%	Sleep disturbance • Sleep initiation: bedtime resistance and sleep anxiety • Sleep maintenance: night wakings Prior interventions • Not reported	High
Bramble[149] Associated publications: Bramble[122] UK	A training curriculum delivered via a single session. Implementation support was delivered via telephone calls	*N* = 15 7.2 years (2.6 years) 67% male All severe learning disability, aetiological factor known for 60%: Down syndrome, 20%; macrocephaly, 7%; Angelman syndrome, 7%; Smith–Magenis syndrome, 7%; carcinuria:, 7%; perinatal cerebral anoxia, 7%; and cerebral leucodystrophy, 7%	Sleep disturbance • Sleep initiation: severe night settling (taking ≥ 1 hour to settle) • Sleep maintenance: night waking problems (most nights of the week and disturbing parents during the night). Sleep problems to be present for at least 1 year Prior interventions • Previous interventions indicated but not described other than sedatives	High

Study details and study design	Intervention details	Participant characteristics: number randomised (*N*), mean age (SD); % male; % diagnosis	Type of sleep disturbance/prior sleep interventions	Risk of bias (low, unclear or high)
Reed *et al.*[129] Associated publications: Reed *et al.*[150] Canada	Group delivery of a training curriculum over three sessions	*N* = 22 5.8 years (2.7 years) 82% male ASD, 100%	Sleep disturbance • Sleep initiation: difficulty falling asleep • Sleep maintenance: night wakings and early morning wakings Prior interventions • Not reported	Unclear
Yu *et al.*[151] Hong Kong	Group delivery of a training curriculum over three sessions, supported by weekly telephone calls. Implementation support was delivered by telephone calls	*N* = 54 4.78 years (0.85 years) 79.6% male ASD, 96%; and Asperger syndrome, 2%	Sleep disturbance • Sleep initiation: bedtime resistance and difficulty initiating sleep (delayed by > 1 hour) • Sleep maintenance: night-time waking that disturbed parents and early wake time (earlier than 06.00) Prior interventions • Not reported	High

continued

TABLE 36 Study details for non-pharmacological interventions (*continued*)

Study details and study design	Intervention details	Participant characteristics: number randomised (*N*), mean age (SD); % male; % diagnosis	Type of sleep disturbance/prior sleep interventions	Risk of bias (low, unclear or high)
Non-comprehensive parent-directed interventions *Cluster trials*				
Wiggs and Stores[152] Associated publications: Wiggs and Stores[153,154] UK	Intervention: a tailored intervention. There was a single session for the assessment and development of a sleep management strategy and training the parent in the strategy. Implementation support was delivered via telephone calls Control: no intervention	*N* = 30 Individually tailored behavioural programme (*n* = 15): • 8.21 years (2.7 years) • 60% male • Unknown, 40%; unknown + autism, 27%; Down syndrome, 7%; meningitis, 7%; microcephaly, 7%; cerebral palsy, 7%; and CHARGE association, 7%. In addition, 33% had epilepsy (absence, 7%; tonic clonic, 13%; absence and tonic clonic, 7%; and atonic, 7%) Control (*n* = 15): • 10.77 years (3.81 years) • 60% male • Unknown, 27%; unknown + autism, 20%; Down syndrome, 20%; agenesis of the corpus callosum, 7%; Sanfillipo syndrome, 7%; Ring 15 chromosome disorder, 7%; and cerebral palsy, 13%. In addition, 40% had epilepsy (absence, 7%; tonic clonic, 13%; tonic, 7%; tonic clonic and complex partial, 7%; and tonic, complex partial and absence, 7%)	Sleep disturbance 'Severe problems' defined as: • Sleep initiation: settling problems occurring ≥ 3 times/week, taking > 1 hour to settle and fall asleep • Sleep maintenance: when parents were disturbed, night waking occurring ≥ 3 times/week; when duration of wake was more than a few minutes and parents were disturbed, waking before 5 a.m. ≥ 3 times/week Prior interventions • Not reported	High

Study details and study design	Intervention details	Participant characteristics: number randomised (*N*), mean age (SD); % male; % diagnosis	Type of sleep disturbance/prior sleep interventions	Risk of bias (low, unclear or high)
Before-and-after study				
Peppers *et al.*[155] USA	Prescriptive sleep hygiene advice delivered in a single session	*N* = 23 Prescriptive sleep hygiene intervention (*n* = 23): • Age: not reported • 57% male • ADHD, 87%; and ADD, 13% Control (*n* = 30): • Age: not reported • Gender: not reported • Diagnosis: not reported	Sleep disturbance • A score of ≥ 42 on the sleep assessment tool (intervention group) • A score of ≤ 41 on the sleep assessment tool (control group)	High
Other non-pharmacological interventions *Parallel trials*				
Piazza *et al.*[157] USA	Intervention: FBRC, for 10 days. The study author delivered the face-to-face (home visits) and booklet intervention Control: bedtime scheduling	*N* = 14 FBRC (*n* = 7): • 6.7 years (2.6 years) • Gender: not reported • All moderate to profound developmental disability. In addition (not mutually exclusive): PDD, 14%; cerebral palsy, 14%; Down syndrome, 14%; Prada-Willi, 14%; sleep apnoea, 14%; mixed motor encephalopathy, 14%; autism, 29%; and seizure disorder, 14% Bedtime scheduling (*n* = 7): • 8.3 years (3.0 years) • Gender: not reported	Sleep disturbance • Sleep maintenance: sleeping for at least 10% less than what is expected for age, including night and early waking • Sleep initiation: delayed sleep onset Prior interventions • Children were excluded if they were receiving pharmacological interventions for sleep. No other previous interventions reported	High

continued

TABLE 36 Study details for non-pharmacological interventions (*continued*)

Study details and study design	Intervention details	Participant characteristics: number randomised (*N*), mean age (SD); % male; % diagnosis	Type of sleep disturbance/prior sleep interventions	Risk of bias (low, unclear or high)
		• All moderate to profound developmental disability. In addition (not mutually exclusive): cerebral palsy, 29%; seizure disorder, 71%; Down syndrome, 14%; and autism, 29%		
Crossover trials				
Francis and Dempster[156] Australia	Intervention: valerian: 500 mg per tablet, 30 mg per kilogram of body weight as a single nightly dose at least 1 hour before bedtime, taken for 2 weeks Control: to give the same appearance and odour, the placebo contained 25 mg of whole room *V. edulis* extract, taken for 2 weeks	*N* = 5 Age range: 7–14 years 100% male Moderate intellectual disability, 40%; mild to moderate intellectual disability, 20%; genetic disorder, 20%; episodic fibril convulsions, 20%; hyperactivity and behaviour problems, 20%; ADD, 20%; and ADHD, 20%	Sleep disturbance 'Significant sleep problems' including: • Sleep initiation: sleep initiation • Sleep maintenance: sleep maintenance, co-sleeping and morning waking Prior interventions • Not reported	High
Gringras *et al.*[36] Associated publications: Gringras *et al.*[36] UK	Intervention: a weighted blanket. The blanket weight was 2.25 kg (small) or 4.5 kg (large). Blankets used for 12–16 days and were given by researchers at home/clinic visits Control: a placebo blanket	*N* = 73 Intervention + control (*n* = 36): • 8.7 years (3.3 years) • 78% male • Autism: 22%; Asperger syndrome: 44%; and ASD: 22% Control and intervention (*n* = 37): • 9.9 years (2.8 years) • 70% male • Autism, 35%; Asperger syndrome, 46%; and ASD, 14%	Sleep disturbance • Sleep initiation: delayed sleep onset and/or • Sleep maintenance: poor sleep maintenance Prior interventions • Not reported	High

Study details and study design	Intervention details	Participant characteristics: number randomised (*N*), mean age (SD); % male; % diagnosis	Type of sleep disturbance/prior sleep interventions	Risk of bias (low, unclear or high)
Before-and-after studies				
Guilleminault *et al.*[158] USA	Light therapy + behavioural programme. Light exposure was given daily at 07.00 and 12.00 for 45 minutes (the overall treatment duration, setting and practitioner unclear)	*N* = 142.9 years, range 9 months to 4 years 57% male Moderate to severe learning disabilities, 100%	Sleep disturbance • Sleep maintenance: nocturnal sleep disturbances • Other: 'lack of sleep consolidation' Prior interventions • Sleep medications not otherwise detailed. Eight children had attended sleep clinics and centres for treatment. Behavioural treatment had been implemented with some of the children	High
Oriel *et al.*[159] A–B–A withdrawal design USA	An aquatic exercise programme. 60 minutes of aquatic exercise was undertaken two times per week	*N* = 8 8.88 years, range 6–11 years 63% male ASD, 100%	Sleep disturbance • Parent/guardian report of sleep dysfunction Prior interventions • Children who had complex medical comorbidity in addition to ASD, or any other developmental disorders were excluded	High

continued

TABLE 36 Study details for non-pharmacological interventions (*continued*)

Study details and study design	Intervention details	Participant characteristics: number randomised (*N*), mean age (SD); % male; % diagnosis	Type of sleep disturbance/prior sleep interventions	Risk of bias (low, unclear or high)
Yehuda *et al.*[160] Controlled before-and-after study Israel	Intervention: essential fatty acids supplement that comprised 90 g of α-linolenic and 360 g of linoleic acid in mineral oil. Two capsules per day for 10 weeks Control: placebo (and healthy control group)	*N* = 78 Essential fatty acid supplement (*n* = 40): • Age range (across intervention and control): 9–12 years • 100% male • ADHD: 100% ADHD control (*n* = 38): • 100% male • ADHD: 100% Healthy control (*n* = 22): • 22% male	Sleep disturbance 'Sleep deprived' Prior interventions Not reported	Unclear
Yu and Hong[161] China	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	*N* = 30 6.9 years (3.1 years) 77% male Learning disabilities, 100%	Sleep disturbance Sleep restlessness including: • Sleep initiation: sleep 'starting', circadian rhythm derangement • Sleep maintenance: sleep 'keeping' • Other: abnormal sleep state (including apnoea) Prior interventions • Not reported	High

ADD, attention deficit disorder; CHARGE, coloboma, heart defects, atresia choanae (also known as choanal atresia), growth retardation, genital abnormalities, and ear abnormalities; FBRC, faded bedtime with response cost; LD, learning disability; PDD, Pervasive Developmental Disorder; PDD-NOS, Pervasive Developmental Disorder Not Otherwise Specified.

Appendix 10 Intervention and control details table: non-pharmacological studies

TABLE 37 Intervention and control details: non-pharmacological studies

Study	Mode of delivery: active arm(s)	Intervention overview: the chronological order of delivery	The intervention period; the duration of each parent–practitioner contact; intensity	Content of the intervention	Group delivery only: were parents supported to apply learning to their own child?	For a specific ND?	Location	Who delivered it?
Tailored interventions *Study objective: evaluation of intervention clinical effectiveness*								
Parallel trials								
Hiscock *et al.*[138]	Individual (face to face and telephone)	Intervention: • 1 × face-to-face session Implementation support: • 1 × face-to-face session • 1 × telephone call: offered 2 weeks after the second face-to-face session Control: • Usual care	Intervention: • Intervention period: 4 weeks • Face-to-face sessions ($n = 2$): duration not reported • Telephone calls ($n = 1$): duration of calls not reported	Intervention: • Session 1: assessment; identification of parent goals; sleep education (normal sleep, sleep cycles and sleep hygiene strategies); formulation of a behavioural sleep management plan tailored to the child's sleep problem • Session 2: review of the sleep diary; reinforcement of the management plan created in Session 1; 'trouble shoot any problems' • Telephone call: review of sleep diary; reinforce management plan created in Session 1; trouble shoot any problems	N/A	ADHD	Paediatrican's office, hospital clinic or home	Psychologist or consultant paediatrician
Johnson *et al.*[107]	Individual (face to face)	Intervention: • 4 × face-to-face sessions Implementation support: • 1 × face-to-face session Control: • Non-sleep-related parent education	Intervention: • Intervention period: unclear • Face-to-face sessions ($n = 5$) at 1–1.5 hours; the spacing of the sessions was not reported Control: • Intervention period: unclear • Face-to-face sessions ($n = 5$) at 1–1.5 hours; the spacing of the sessions was not reported	Intervention: • Session 1: basic behavioural principles • Session 2: addressing prevention techniques and bedtime routines • Session 3: addressing reinforcement and extinction procedures for bedtime struggles, night awakenings and early morning awakenings • Session 4: addressing delayed sleep onset and sleep association procedures • Session 5: booster and maintenance session	N/A	ADHD	Clinic	Masters-level doctoral students or senior behavioural analyst

Study	Mode of delivery: active arm(s)	Intervention overview: the chronological order of delivery	The intervention period; the duration of each parent–practitioner contact; intensity	Content of the intervention	Group delivery only: were parents supported to apply learning to their own child?	For a specific ND?	Location	Who delivered it?
				Session content tailored to the family's specific needs and 'a cumulative bedtime and sleep intervention plan developed', modified as needed over the intervention period [A detailed overview of the sessions and content of the control were provided in the paper]				
Moss *et al.*[124]	Group and individual (face to face and telephone)	Intervention:[232] • 2 × workshops (group size 5–7) Published as *The Sleepwise Program*[232] • 1 × face-to-face session Implementation support: • 1 × face-to-face session • Telephone calls provided on a needs basis Control: • Waiting list	Intervention: • Intervention period: unclear • Workshops ($n = 2$) at 3 hours • Face-to-face sessions ($n = 2$): duration not reported • Telephone calls: duration of calls not reported. Provided on a 'needs basis'	Intervention: • Workshop 1: on typical sleep, the nature of sleep and factors affecting sleep, positive sleep practices, types of sleep disturbance; a sleep diary was completed for homework • Workshop 2: on managing sleep disturbance and developing a sleep plan • Face-to-face session 1: sleep interview (i.e. assessment), observation of child's sleep environment and problem-solving for the sleep plan; the sleep plan was produced • Face-to-face session 2: implementation of sleep plan • Telephone calls: ongoing support regarding implementation of sleep plan *[A detailed overview of workshop content is provided in the paper]*	N/A	No	Workshop: not stated Face-to-face sessions: home	Allied health workers

continued

TABLE 37 Intervention and control details: non-pharmacological studies (*continued*)

Study	Mode of delivery: active arm(s)	Intervention overview: the chronological order of delivery	The intervention period; the duration of each parent–practitioner contact; intensity	Content of the intervention	Group delivery only: were parents supported to apply learning to their own child?	For a specific ND?	Location	Who delivered it?
Before-and-after studies								
Austin *et al.*[123]	Group and individual (face to face and telephone)	Intervention:[232] • Published as *The Sleepwise Program*[232] • 2 × workshops (group size 6) • 1 × face-to-face session • 1 × workshop • Implementation support: weekly telephone calls	Intervention period: 15 weeks Workshops ($n = 3$) at 3 hours each Face-to-face sessions ($n = 1$): at ≈2 hours (Workshops 1 and 2 were 2 weeks apart; the face-to-face session was a week later; and workshop 3 was a week after that) Telephone calls: duration of calls not reported; delivered weekly for 6 weeks	• Workshop 1: on typical sleep, the development of sleep, sleep cycles, nature of sleep, factors influencing children's sleep patterns, need for sleep and positive sleep practices • Workshop 2: on causes of sleep disturbance, prevalence of sleep disturbance, types of sleep disturbance disorders and preparation for starting a sleep plan • Face-to-face session 1: sleep interview (i.e. comprehensive sleep assessment) and observation of child's bedroom environment • Workshop 3: positive sleep practices, establishing a bedtime routine, sensory, behavioural and communication approaches, positive reinforcement, goal-setting and the development of a sleep plan • Telephone calls: provide ongoing support as parents implement the sleep plan	N/A	No	Workshops: not stated Face-to-face session: home	Psychologist or manager of early intervention centre
Beresford *et al.*[21]	Individual (face to face)	2 × face-to-face sessions Implementation support: face-to-face sessions – number determined by progress	Intervention period: variable, usually 12–16 weeks Face-to-face sessions ($n =$ variable): duration not reported	• Sessions 1 and 2: assessment, home visit sleep strategy created and parent education and training in sleep strategy • Sessions 3 onwards: supporting implementation of sleep strategy The number of sessions is directed by progress towards the outcomes	N/A	No	Community centre and home	Community-based disability link workers

Study	Mode of delivery: active arm(s)	Intervention overview: the chronological order of delivery	The intervention period; the duration of each parent–practitioner contact; intensity	Content of the intervention	Group delivery only: were parents supported to apply learning to their own child?	For a specific ND?	Location	Who delivered it?
Quine and Wade[146]	Individual (face to face)	2 × face-to-face sessions Implementation support: face-to-face sessions – number determined by progress	Intervention period: variable, 6–28 weeks Face-to-face sessions (n = variable): duration not reported. They took place weekly, but after session 3, the authors sought to increase the spacing of the sessions	• Session 1: assessment and parent education • Session 2: (sleep) training programme devised and parents trained in implementation • Session 3 onwards: supporting the implementation of the sleep strategy The number of sessions is directed by progress towards the outcomes	N/A	No	Home	Health professional: health visitor, community nurse or district nurse, school nurse
Weiksop *et al.*[126]	Individual (face to face and telephone)	4 × face-to-face sessions Implementation support: 1 × face-to-face session Telephone calls weekly after session 4, diminishing in intensity after session 5	Intervention period: minimum 7 weeks Face-to-face session 1: duration not reported Face-to-face sessions 2–4: duration not reported; consecutive weeks Face-to-face session 5: duration not reported; delivered 5 weeks after session 4	• Session 1: assessment • Session 2: goal-setting, parent education and creation of sleep hygiene/bedtime routine strategy • Session 3: parent education (supporting consistent parenting and positive problem-solving) • Session 4: training in three extinction techniques, and the parent chooses the specific technique that they will implement – further training is given in chosen technique • Session 5: review of progress	N/A	No	Home and clinic	'Therapist'

continued

TABLE 37 Intervention and control details: non-pharmacological studies (*continued*)

Study	Mode of delivery: active arm(s)	Intervention overview: the chronological order of delivery	The intervention period; the duration of each parent–practitioner contact; intensity	Content of the intervention	Group delivery only: were parents supported to apply learning to their own child?	For a specific ND?	Location	Who delivered it?
Study objective: comparative evaluation of modes of delivering implementation support [home visit (arm a) vs. telephone call (arm b)]								
Parallel trial								
Beresford *et al.*[21]	Arm A: individual (face to face) Arm B: individual (face to face and telephone)	Arm A: • 1 × face-to-face session • Implementation support: face-to-face sessions approximately weekly over a 6- to 8-week period; number determined by progress Arm B: • 1 × face-to-face session • Implementation support: telephone calls approximately weekly	Arm A: • Intervention period: variable, ≈10 weeks • Face-to-face sessions ($n = 1$ + variable): duration of sessions not reported; implementation support sessions were delivered approximately weekly, over a 6- to 8-week period Arm B: • Intervention period: variable, ≈10 weeks • Face-to-face session ($n = 1$): duration of session not reported • Telephone calls (n = variable): duration of calls not reported; delivered approximately weekly, over a 6- to 8-week period	• Session 1 (arms A and B): comprehensive sleep assessment, development of sleep management strategy and parent education and training in strategy • Sessions 2 onwards (arm A only): support with implementation of sleep strategy • Telephone calls (arm B only): support with implementation of sleep strategy	N/A	No	Home	Specialist health visitors

Study	Mode of delivery: active arm(s)	Intervention overview: the chronological order of delivery	The intervention period; the duration of each parent–practitioner contact; intensity	Content of the intervention	Group delivery only: were parents supported to apply learning to their own child?	For a specific ND?	Location	Who delivered it?
Study objective: comparative evaluation of intervention intensity (brief vs. extended)								
Parallel trial								
Sciberras *et al.*[143]	Brief intervention: individual (face to face) Extended intervention: individual (face to face and telephone)	Brief intervention: • 1 × face-to-face session Extended intervention: • 2 × face-to-face sessions Implementation support: • 1 × telephone call • Further face-to-face session if required	Brief intervention: • Intervention period: a single face-to-face session • Face-to-face session ($n = 1$) at 45 minutes Extended intervention: • Intervention period: 4 weeks • Face-to-face sessions ($n = 2$) at 45 minutes, 1 week apart • Telephone call ($n = 1$): duration not reported; delivered 2 weeks later	Brief and extended intervention: • Described as 'a behavioral sleep program'[143] (p. 933). The content of the written material used and developed to supplement sessions suggests that the face-to-face session(s) covered: 'normal sleep, healthy sleep hygiene practices and standard clinical strategies for managing sleep problems'[143] (p. 933) Extended only: • Telephone call: described as a 'follow-up call'[143] (p. 933)	N/A	ADHD	Hospital	Paediatric trainee or child psychologist
Non-tailored interventions *Study objective: evaluation of intervention effectiveness*								
Parallel trial								
Adkins *et al.*[127]	Written material	Intervention: • Provision of a 'sleep education pamphlet' to parents (four pages) Implementation support: • None Control: • No pamphlet	N/A	Intervention: • The topics covered in the pamphlet were 'providing a comfortable sleep setting; establishing regular bedtime habits; keeping a regular schedule; teaching your child to fall asleep alone; avoiding naps (in children who have outgrown the need for a daytime nap); encouraging daytime activities that promote a better sleep/wake schedule.'[127] (p. S140)	N/A	ADHD	N/A	N/A

continued

TABLE 37 Intervention and control details: non-pharmacological studies (*continued*)

Study	Mode of delivery: active arm(s)	Intervention overview: the chronological order of delivery	The intervention period; the duration of each parent–practitioner contact; intensity	Content of the intervention	Group delivery only: were parents supported to apply learning to their own child?	For a specific ND?	Location	Who delivered it?
Before-and-after studies								
Beresford *et al.*[21]	Group	4 × workshops (maximum group size 8) Implementation support: none except that included in workshop curriculum	Intervention period: 5 weeks Workshops ($n = 4$) at 3 hours; delivered over a 5-week period	Workshop 1: 'children's sleep; behavioural approaches to behaviour management; positive and negative reinforcers; communication'[21] Workshop 2: 'sleep routines; structuring bedtime; using reinforcers to manage behaviour; planning bedtime routines: bedroom environment'[21] Workshop 3: 'principles of behavioural analysis then applied to children's sleep problems'[21] Workshop 4: 'specific strategies to manage sleep problem behaviours; the use of medication'[21]	Yes	No	Community setting	Staff based in Learning Disability Child Mental Health Service
Beresford *et al.*[21]	Group	1 × workshop (maximum group size 20).[233] (www.scope.org.uk/support/services-directory/sleep-solutions-training-for-families; accessed 20 April 2016) Implementation support: none	Intervention period: a single face-to-face session Face-to-face session ($n = 1$) at 5 hours	Workshop topics: 'the impact of sleep disorders; your existing routine; the bedroom environment; does your child's impairment matter?; techniques, tips and resources; making a change, when the time is right'[21]	No	No	Community setting	Staff trained in managing sleep disturbance and working for Scope UK (a UK charity for children with cerebral palsy and their families)
Bramble[149]	Individual (face to face and telephone)	1 × face-to-face session Implementation support: telephone calls delivered on 3 consecutive days after the face-to-face session, with additional calls arranged, if necessary, for up to 2 weeks	Intervention period: up to 2 weeks Face-to-face session ($n = 1$): duration not reported Telephone calls ($n =$ variable): duration not reported. Delivered 3 consecutive days after face-to-face session, with additional calls arranged if necessary for up to 2 weeks	Session 1: education in children's sleep and behaviour modification based on the following principles: 'setting regular bed and wake times; establishing routines; setting appropriate mood for sleep; rapid settling of child at bedtime; parental withdrawal after settling; ignoring protestations; returning child to room with minimal contact; rewarding and praising improved morning and night-time behaviour'[149] (p. 544) The content of the session was subject to minimal adaptation only in specific cases	N/A	No	Clinic or home	Consultant psychiatrist

Study	Mode of delivery: active arm(s)	Intervention overview: the chronological order of delivery	The intervention period; the duration of each parent–practitioner contact; intensity	Content of the intervention	Group delivery only: were parents supported to apply learning to their own child?	For a specific ND?	Location	Who delivered it?
				Telephone calls: provided support to parents as they changed the ways that they managed sleep disturbance				
Reed *et al.*[129]	Group	3 × workshops (group size 3–5) Implementation support: none except that included in workshop curriculum	Intervention period: 3 weeks Workshops ($n = 3$): at 2 hours; delivered over consecutive weeks	Workshop 1: establishing daytime and night-time habits and the child's bedtime routine Workshop 2: minimising night wakings and early morning awakenings Workshop 3: addressing individualized sleep concerns[129] (pp. 939–40) *[A detailed overview of the workshop content is provided in the paper]*	Yes	ASD	Not reported	Neurology sleep specialist, with assistance from a nurse educator and educational consultant
Yu *et al.*[151]	Group and individual (telephone)	3 × workshops (maximum group size 8) Implementation support: telephone calls weekly for the entire intervention period	Intervention period: 7 weeks Workshops ($n = 3$): duration not reported; delivered over consecutive weeks Telephone calls ($n = 7$): weekly for entire intervention period; duration of calls not reported	Workshop 1: ASD and sleep; impacts; sleep hygiene Workshop 2: promoting sleep – behavioural approaches Workshop 3: other concerns; sleep medication for sleep problems Telephone calls: offer parents immediate support post workshop, answer enquiries and gain feedback	Yes	ASD	Not reported	Trained research nurse

continued

TABLE 37 Intervention and control details: non-pharmacological studies (*continued*)

Study	Mode of delivery: active arm(s)	Intervention overview: the chronological order of delivery	The intervention period; the duration of each parent–practitioner contact; intensity	Content of the intervention	Group delivery only: were parents supported to apply learning to their own child?	For a specific ND?	Location	Who delivered it?
Study objective: comparative evaluation of mode of intervention delivery vs. no intervention								
Parallel trial								
Montgomery *et al.*[49]	Arm A: written material Arm B: individual (face to face)	Arm A: • Provision of a 14-page easy-to-read, illustrated booklet • Implementation support: none Arm B: • 1 × face-to-face session delivering same material as covered in booked (conventional approach) • Implementation support: none Arm C (control): • No intervention	Arm A: • Intervention period: N/A Arm B: • Intervention period: a single face-to-face session • Face-to-face session ($n = 1$): at 90 minutes	Arms A and B: • Topics covered: 'normal sleep; introduction to behavioural techniques in general; monitoring behaviour; good sleep habits; specific techniques for changing undesirable behaviour'[49] (p. 126) *[A detailed overview of content provided in paper]*	N/A	No	Arm A: • Home Arm B: • Home	Arms A and B: • Member of research team

Study	Mode of delivery: active arm(s)	Intervention overview: the chronological order of delivery	The intervention period; the duration of each parent–practitioner contact; intensity	Content of the intervention	Group delivery only: were parents supported to apply learning to their own child?	For a specific ND?	Location	Who delivered it?
Study objective: comparative evaluation of alternative modes of intervention delivery								
Parallel trial								
Malow *et al.*[128]	Arm A: group and individual (telephone) Arm B: individual (face to face and telephone)	Arm A: • 1 × workshop (group size 2–4) • 1 × telephone call • 1 × workshop • 1 × telephone call • Implementation support: telephone calls – delivered after each workshop Arm B: • 1 × face-to-face session • Implementation support: telephone calls: within 2 weeks of receiving the face-to-face session	Arm A: • Intervention period: unclear • Workshops ($n = 2$): at 2 hours • Telephone calls ($n = 2$): duration not reported; calls delivered after each workshop Arm B: • Intervention period: 2 weeks • Face-to-face session ($n = 1$) at 1 hour • Telephone calls ($n = 2$): duration not reported; delivered within 2 weeks of face-to-face session and 1 week apart	Arms A and B: • 'Major areas covered in sessions were: sleep hygiene (including daytime and evening habits and the sleep environment); sleep amount/timing/regularity; bedtime routine; strategies related to minimizing bedtime resistance, night wakings, co-sleeping (including graduated extinction, bedtime pass)'[128] (pp. 219–220) • Telephone calls: described as 'educational phone calls . . . to review questions and answer any questions parents may have'[128] (p. 220) *[A detailed overview of content provided in paper. Same curriculum as Reed* et al.[129] *but aspects of intervention delivery differ]*	Group delivery only: were parents supported to apply learning to their own child?': Yes	ADHD	Arm A: • Not reported Arm B: • Not reported	Arms A and B: • 'Trained educators'

continued

TABLE 37 Intervention and control details: non-pharmacological studies (*continued*)

Study	Mode of delivery: active arm(s)	Intervention overview: the chronological order of delivery	The intervention period; the duration of each parent–practitioner contact; intensity	Content of the intervention	Group delivery only: were parents supported to apply learning to their own child?	For a specific ND?	Location	Who delivered it?
Non-comprehensive parent-directed interventions								
Tailored intervention: content restricted to behavioural principles of managing problem behaviour *Study objective: evaluation of intervention clinical effectiveness (intervention vs. attention control)*								
Parallel trial								
Wiggs and Stores[152]	Intervention: individual (face to face and telephone) Attention control: individual (face to face)	Intervention: • 1 × face-to-face session • Implementation support: telephone calls at least weekly Attention control: • 1 × face-to-face session • Implementation support: none	Intervention: • Intervention period: variable, up to 3 months • Face-to-face session ($n = 1$) at 1–2.5 hours • Telephone calls: • (n = variable); at least weekly, and for up to 3 months • Duration of call not reported Attention control: • Intervention period: single face-to-face session • Face-to-face session ($n = 1$) at 1–2.5 hours	Intervention: • Face-to-face session: 'functional assessment of problem; identification of parents' aims of treatment, discussion of possible mechanisms maintaining settling and waking problems; discussion of therapeutic techniques . . . practical application; identification and anticipation of particular problems with intervention; identification of targets for first stage'[152] • Telephone calls: to monitor progress, encourage, discuss any problems and amend programme as necessary. Parents also told could telephone researcher at any time (this rarely happened)'[152] (p. 121) Attention control: • Content not described	N/A	No	Both arms: • Home	'The researcher'

Study	Mode of delivery: active arm(s)	Intervention overview: the chronological order of delivery	The intervention period; the duration of each parent–practitioner contact; intensity	Content of the intervention	Group delivery only: were parents supported to apply learning to their own child?	For a specific ND?	Location	Who delivered it?
Tailored intervention: content restricted to sleep hygiene								
Before-and-after study								
Peppers[155]	Individual (face to face)	Intervention: • 1 × face-to-face session • Implementation support: none	Intervention period: a single face-to-face session Face-to-face session ($n = 1$): duration not reported	Session content: 1. The parent/caregiver and child view 6-minute standardised sleep hygiene video (see p. e44 for content) 2. Opportunity to discuss the video and key aspects of the sleep hygiene routine with practitioner 3. Development of a patient specific sleep hygiene routine, then embedded in the electronic health record for documentation 4. Provision of a written copy of the patient specific sleep hygiene routine'[155] (p. e46)	N/A	ADHD	Clinic	Physician or nurse practitioner
Other non-pharmacological interventions								
Parallel trials								
Piazza *et al.*[157]	Individual (face to face)	Intervention: • Faded bedtime and response costs Control: • Bedtime scheduling	'Average treatment length 8 weeks'	Faded bedtime with response cost involved establishing a bedtime in which it was likely that the child would fall asleep within 15 minutes. Response cost involved keeping the child awake for 1 hour if they did not fall asleep within 15 minutes of bedtime Bedtime reduced by half an hour each night, if the child fell asleep within 15 minutes of bedtime the previous night. Bedtime increased by half an hour if the child did not fall asleep within 15 minutes the previous night	N/A	No	Hospital	Not specified

continued

TABLE 37 Intervention and control details: non-pharmacological studies (*continued*)

Study	Mode of delivery: active arm(s)	Intervention overview: the chronological order of delivery	The intervention period; the duration of each parent–practitioner contact; intensity	Content of the intervention	Group delivery only: were parents supported to apply learning to their own child?	For a specific ND?	Location	Who delivered it?
Crossover trials								
Francis and Dempster[156]	Individual (face to face)	Intervention: • Valerian tablet Control: • Placebo tablet	Treatment A: 2 weeks Washout period 1: 1 week Treatment B: 2 weeks Washout period 2: 1 week	Intervention: • Valerian tablet. 500 mg per tablet. 30 mg per kilogram of body weight as a single nightly dose 1 hour before bedtime, taken for 2 weeks Control: • To give the same appearance and odour, the placebo tablet contained 25 mg of whole room *V. edulis* extract	N/A	No	Tablets administered at home	Parent administered tablets to child
Gringas *et al.*[36]	Not explicitly stated, reasons for using weighted blankets 'the deep pressure and more consistent sensory input provided by weighted items reduces the body's physiological level of arousal and stress, which might improve sleep'	Intervention: • Weighted blanket and additional clinic/home visit Control: • Placebo blanket	At baseline, participants received a weighted blanket, which was used for 12–16 days At 2-week follow-up, the initial blanket was removed and the alternative blanket was provided and used for 12–16 days	Intervention: • During a home or clinic visit, children were given a weighted blanket at baseline and it was used for 12–16 days. Blankets weighed 2.25 kg (small) or 4.5 kg (large). Additional clinic/home visit Control: • During a home or clinic visit, children were given a control blanket at baseline and it was used for 12–16 days. Blankets weighed 2.25 kg (small) or 4.5 kg (large)	N/A	No	Blankets received at home or clinic Visits were at home or in a clinic. Blankets used at child's home	Researchers
Before-and-after studies								
Guilleminault *et al.*[158]	Individual	Intervention: • Light therapy and behavioural programme	Overall treatment duration: unclear Light exposure duration: daily at 7 a.m. and 12 a.m. for 45 minutes	Light therapy and behavioural programme Children were exposed to bright light (sunlight or artificial) A behavioural programme was also implemented and involved scheduled parent child interaction, scheduled naps for younger children, avoidance of naps for older children, scheduled lunch and scheduled sleep time	N/A	No	Unclear	Unclear

Study	Mode of delivery: active arm(s)	Intervention overview: the chronological order of delivery	The intervention period; the duration of each parent–practitioner contact; intensity	Content of the intervention	Group delivery only: were parents supported to apply learning to their own child?	For a specific ND?	Location	Who delivered it?
Oriel *et al.*[159]	Group	Intervention: • Aquatic exercise programme	Intervention: 60 minutes of aquatic exercise two times per week	Aquatic exercise programme: • During all three phases of the study, the researchers made telephone calls to the parents/guardians questioning them about their child's previous night of sleep (two times per 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	No	No	Not specified	It is not clear who delivered exercise programme – each participant was paired with a researcher or volunteer
Yehuda *et al.*[160]	Individual	Intervention: • Essential fatty acids supplement Control: • placebo (and a healthy control group)	Two capsules per day for 10 weeks	Intervention: • Essential fatty acid supplement, which comprised 90 g of α-linolenic and 360 g of linoleic acid in mineral oil Control: • A placebo, which was mineral oil in an identical capsule	No	No	Not specified	Not specified
Yu and Hong[161]	Individual	Intervention: • 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	See full paper for technical details of acupuncture and ear-point taping	No	No	Not specified	Not specified

N/A, not applicable.

Appendix 11 Child-related outcomes for studies evaluating parent-directed tailored interventions

TABLE 38 Child-related outcomes for studies evaluating parent-directed tailored interventions

Study	Child sleep-related outcome assessed	Method of assessment	Definitions[a]
Global measures and composite scores			
Austin *et al.*[123]	CSHQ	CSHQ (total score)	The CSHQ was used to measure overall total sleep disturbance in children aged 4–12 years. Subscales include bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night wakings, parasomnias, sleep disordered breathing and daytime sleepiness. There were 33 items rated on a three-point Likert scale. The authors provide a reference
Beresford *et al.*[21]	CSHQ	CSHQ (total score and subscales)	The CSHQ was used to assess the severity of sleep problems in children aged 4–10 years. Parent reported. Subscales include bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night wakings, parasomnias, sleep disordered breathing and daytime sleepiness. The authors provide a reference
	Parent-set child sleep goal(s)	Parent-set child sleep goal(s)	A 10-point scale captured progress towards goals. Parents identified up to three goals during session 1. The authors provide a reference
Beresford *et al.*[21]	CSHQ	CSHQ (total score and subscales)	The CSHQ was used to assess the severity of sleep problems in children aged 4–10 years. Parent reported. Subscales include bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night wakings, parasomnias, sleep disordered breathing and daytime sleepiness. The authors provide a reference
	Parent-set child sleep goal(s)	Parent-set child sleep goals	A 10-point scale captured progress towards goals. Parents identified up to three goals during session 1. The authors provide a reference
Hiscock *et al.*[138]	CSHQ	CSHQ (total score)	The CSHQ was used, a validated 33-item parent-reported measure of difficulties initiating and maintaining sleep over the past week. The maximum total score was 99. The authors provide a reference
	Sleep efficiency	Actigraphy	The ratio of time asleep to time spent in bed
	TST	Actigraphy	The duration of night-time sleep
Johnson, *et al.*[107]	Composite sleep index (bedtime resistance, night waking, early waking, sleeping in places other than bed)	Composite sleep index of the modified version of the Simonds and Parraga Sleep Questionnaire	A modified version of the Simon and Parraga Sleep Questionnaire completed by child's primary caregiver. The Total Composite Sleep Index score ranged from 0 to 12. It was calculated by assigning a score to the frequency of targeted sleep problems: bedtime resistance, night awakening, early awakening and sleeping in places other than bed. Additional scores were assigned to the duration of sleep latency and night awakenings. To calculate frequency, scores were as follows: 1 = problems occurring once or twice a week and 2 = problems occurring more than several times a week. To calculate duration, scores were as follows: 1 = sleep latency lasting up to 1 hour and 2 = over 1 hour. For night awakenings, scores were as follows: 1 = awakenings lasting a few minutes and 2 = lasted longer than a few minutes. The authors provide a reference for the Simond and Parraga Sleep Questionnaire

continued

TABLE 38 Child-related outcomes for studies evaluating parent-directed tailored interventions (*continued*)

Study	Child sleep-related outcome assessed	Method of assessment	Definitions[a]
	Sleep efficiency	Actigraphy and sleep diary (to verify actigraphy)	Percentage of time sleeping while in bed and lights off
	TST	Actigraphy and sleep diary (to verify actigraphy)	No definition provided
Moss *et al.*[124]	CSHQ	CSHQ total score and subscales	'A comprehensive parent-report sleep screening instrument designed for school-aged children' was used to identify behaviourally and medically based sleep problems. It consists of a total score and eight subscale scores. The CSHQ includes additional questions relating to hours of daily sleep and length of night waking. Participants only completed the questions that were used to calculate total and subscale scores. The authors provide a reference
	Sleep goals	Goal Attainment Scale	Families defined individual child sleep goals in conjunction with the facilitator to obtain an objective measure of sleep disturbance change. There were five levels of possible outcome levels: 0%, 25%, 50%, 75% and 100%. The child's current sleep pattern was outlined as 0%. 'For example: If the child did not settle to sleep in less than 30 minutes on any night in the week, a parent report of 100% level of success would have been achieved when the child settled to sleep in less than 30 minutes, 6 nights a week.' No reference was provided
Sciberras *et al.*[125]	CSHQ	CSHQ	The CSHQ was used to measure caregiver report of sleep problems and three items from the CSHQ were used to screen children for sleep apnoea. The authors provide a reference but no further details are provided
Weiskop *et al.*[126]	TST	Sleep diary	The average duration of night-time sleep per week. The length of night wakings was subtracted if these data were available
	Child's sleep behaviour goals	GAS	The GAS was used to assess the clinical significance of any changes in child's sleep behaviour and to provide a quantifiable measure of intervention success. The GAS was based on parent-stated goals: a separate GAS was developed for each sleep behaviour identified by parents as a goal for behaviour change. For each behaviour, 0% success was set as the baseline rate of that behaviour. The parents and therapists decided what total success (100%) meant before the intervention. This did not have to mean elimination of the sleep problem but related to the level of improvement that the parents thought would make a difference to their lives and was developmentally appropriate. Post intervention, the change in each behaviour was expressed as a percentage of success over baseline. The authors provide a reference
Sleep initiation			
Austin *et al.*[123]	Bedtime settling: bedtime resistance, bedtime routine and SOL	Sleep diary	No definition provided
Johnson *et al.*[107]	Bedtime settling: SOL	Actigraphy, sleep diary (to verify actigraphy)	Time from lights off to sleep onset

TABLE 38 Child-related outcomes for studies evaluating parent-directed tailored interventions (*continued*)

Study	Child sleep-related outcome assessed	Method of assessment	Definitions[a]
Quine and Wade[146]	Bedtime settling: child settling	Parent report – Behaviour Screening Questionnaire	Settling was assessed using the behaviour screening questionnaire. Severe settling problem = ≥ 3 times per week. Mild settling problem = once or twice a week. The authors provide a reference for the Behaviour Screening Questionnaire and a detailed description of the questionnaire is provided in an appendix to the Quine and Wade report
Weiskop *et al.*[126]	Bedtime settling:		
	Number of pre-sleep disturbances/ week	Sleep diary	Pre-sleep disturbance was defined as any disruption occurring between the time that the child was put to bed and the time of sleep onset, for example crying, leaving the room or calling out
	Number of nights that child fell asleep in own bed	Sleep diary	No further definition provided
	SOL	Sleep diary	Average sleep latency per week. Sleep latency was considered as the number of minutes between first being settled to bed and sleep onset
Sleep maintenance			
Austin *et al.*[123]	Night waking	Sleep diary	No definition provided
	Waking time	Sleep diary	No definition provided
	Non-specified sleep disturbance: co-sleeping and severity of child's sleep disturbance	Sleep disturbance index	A parent-reported scale of 0–2 with a total maximum score of 8. Parents reported on the severity of their child's sleep. The index focusses on difficulties settling the child to sleep, night-time wakening, parent's attendance to the child during the night and parental sleep loss through co-sleeping
Hiscock *et al.*[138]	Night waking: WASO		
		Actigraphy	No definition was provided
	Non-specified night-time sleep disturbance: sleep problems	Primary caregiver report of child sleep problems over past 4 weeks	Sleep problems were rated as none, mild, moderate or severe
Quine and Wade[146]	Waking time: number of night wakings and sleeping in parent's bed	Parent reported-behaviour screening questionnaire	It was classified as a severe waking problem if it occurs > 3 times per week and the child wakes for more than a few minutes, disturbs parents or goes into the parents' room or bed. The authors provide a reference for the behaviour screening questionnaire and a detailed description of the questionnaire is provided in an appendix to the Quine and Wade report
Sciberras *et al.*[125]	Non-specified night-time sleep disturbance: sleep problems	CSHQ and caregiver report	The CSHQ was used to measure care-giver report of sleep problems and three items from the CSHQ were used to screen children for sleep apnoea. The authors provide a reference but no further details are provided

continued

TABLE 38 Child-related outcomes for studies evaluating parent-directed tailored interventions (*continued*)

Study	Child sleep-related outcome assessed	Method of assessment	Definitions[a]
Weiskop *et al.*[126]	Night waking: Number of night wakings	Sleep diary	Number of night wakings per week that parents were aware of
	Non-specified night-time sleep disturbance: number of nights/week that child co-slept	Sleep diary	Co-sleeping was not coded when the parents only lay with the child until they fell asleep at bedtime
Sleep scheduling			
Austin *et al.*[123]	Napping	Sleep diary	No definition provided
Child-related quality of life, daytime behaviour and cognition			
Austin *et al.*[123]	Behavioural and emotional disturbance	Developmental Behaviour Checklist – parent version	Used to assess behavioural and emotional disturbance in children with developmental and intellectual disabilities. 96 items are grouped into the following subscales: disruptive/antisocial, self-absorbed, communication disturbance, anxiety and social relating. Subscales are combined to determine a total behaviour problem score. The authors provide a reference
Hiscock *et al.*[138]	Child-related quality of life	PedsQL	The PedsQL version 4.0 was used for parent proxy report. It is a validated 23-item measure of quality of life for children aged 2–18 years. Items were rated on a 5-point scale. 15 items contributed to a psychosocial health summary score. Scores ranged from 0 to 100. The authors provide a reference
	Child daytime behaviour and cognition		
	ADHD symptoms	ADHD Rating Scale IV (parent and teacher reported)	The ADHD Rating Scale IV parent- and teacher-reported versions. It is a validated 18-item measure of ADHD symptoms rated on 4-point scale. Nine items assess inattentive symptoms and nine assess hyperactive symptoms. The authors provide a reference
	Daily functioning	Daily parent rating of evening and morning behaviour	A 11-item parent-reported measure of core ADHD symptoms and behavioural problems on a 4-point scale. Scores ranged from 0 to 33. The authors provide a reference
	Behaviour	Strengths and difficulties questionnaire	Parent and teacher versions. It is a validated 25-item measure of behavioural and emotional problems for children aged 4–16 years. Items rated on 3-point scale, with 20-item total problem score from 0 to 40. The authors provide a reference
	Working memory	Working memory test battery for children	Three subtests from the working memory test battery for children assessing the central executive working memory domain: backwards digital recall, counting recall and listening recall. These subsets provide a central executive composite. The authors provide a reference
Moss *et al.*[124]	Child daytime behaviour and cognition	Developmental Behaviour Checklist – Parent Version	Developmental Behaviour Checklist is used to measure the behavioural and emotional problems of children with developmental and intellectual disabilities aged 4–18 years. The checklist includes five subscales: disruptive/antisocial, self-absorbed, communication disturbance, anxiety and societal relating and a total score. The authors provide a reference

TABLE 38 Child-related outcomes for studies evaluating parent-directed tailored interventions (*continued*)

Study	Child sleep-related outcome assessed	Method of assessment	Definitions[a]
Quine and Wade[146]	Child daytime behaviour and cognition	Daytime behaviour (BPI)	The BPI is an adaptation of the Behaviour Screening Questionnaire that includes a wider range of sleep problems that are deemed more appropriate for children with severe learning difficulties. A reference is provided as is a more detailed description of the Index and the Behaviour Screening Questionnaire in an appendix to the report
Sciberras *et al.*[125]	Child related quality of life	PedsQL	Health-related quality of life was measured using PedsQL (version 4.0), psychosocial health summary score. The authors provide a reference
	Child daytime behaviour and cognition	ADHD Rating Scale IV	ADHD symptom severity was measured using the ADHD Rating Scale IV. The authors provide a reference
	Daily parent rating on the Evening and Morning Behaviour Scale	The Evening and Morning Behaviour Scale	Daily functioning was measured via daily parent rating on the Evening and Morning Behaviour Scale. The authors provide a reference
	School attendance	Number of days missed or late for school over previous 6 months	No further definition provided. The authors provide a reference
Other child-related sleep outcomes			
Austin *et al.*[123]	Meal times	Sleep diary	No definition provided
Hiscock *et al.*[138]	School attendance	Parent report of child's school attendance	Parent report of whether or not their child had missed or been late for school over the preceding 3 months
	Sleep help	Parent report of other professional help sought for their child's sleep – for example, general practitioner or psychologist	Parent report of other professional help sought for their child's sleep

GAS, Goal Attainment Score.
a As defined by study authors.

Appendix 12 Parent sleep-related outcomes for studies evaluating parent-directed tailored interventions

TABLE 39 Parent sleep-related outcomes for studies evaluating parent-directed tailored interventions

Study	Parent sleep-related outcome assessed	Method of assessment	Definitions[a]
Parent-carer-related quality of life			
Hiscock *et al.*[138]	Parental stress	DASS	Validated 21-item measure of adult mental health including scales assessing depression, anxiety and stress. Items rated on a 4-point scale. The authors provide a reference
Moss *et al.*[124]	Parent stress	Parenting Stress Index – Short Form	The Parenting Stress Index enables a clinician or researcher to examine the relationship of parenting stress with child characteristics, parent characteristics and situations that are directly related to the role of being a parent. The index includes three subscales: parental distress, parent–child dysfunctional interaction and difficult child. A total stress score is also provided. The authors provide a reference
Quine and Wade[146]	Parent stress	Maternal stress and morale – the Malaise Inventory	A 24-item binary choice questionnaire adapted from the Cornell Medical Index. Scores of 5 or 6 were outside the normal range and indicate stress. Scores of ≥ 7 were considered more critical. The authors provide a reference
Sciberras *et al.*[125]	Mental health	DASS	Mental health was measured via the DASS. The authors provide a reference
Other outcomes			
Hiscock *et al.*[138]	Family functioning	Parent-reported missed work attendance	Parent report of whether or not they had missed or been late for work over the preceding 3 months and the number of days that they missed or were late for work during that period
Sciberras *et al.*[125]	Parent work attendance	Parent/caregiver report	Number of days missed or late to work over the previous 6 months. The authors provide a reference

a As defined by study authors.

Appendix 13 Measures of perceived confidence and/or efficacy and/or understanding of sleep/sleep management for studies evaluating parent-directed tailored interventions

TABLE 40 Measures of perceived confidence and/or efficacy and/or understanding of sleep/sleep management for studies evaluating parent-directed tailored interventions

Study	Method of assessment	Definition[a]
Behavioural		
Beresford *et al.*[21]	PSOC scale and satisfaction and efficacy subscales	A 16-item scale with two subscales. Parents respond to a series of questions about parenting, indicating their level of agreement or disagreement on a six-point Likert scale. The satisfaction subscale measures the extent that parents are satisfied with their role as a parent and so captures the affective dimension of parenting competence, including the extent of parental frustration, anxiety and motivation. The efficacy subscale measures the extent that parents feel that they are managing the role of being a parent and captures competence, problem-solving ability and capability in the parenting role. The authors provide a reference
Beresford *et al.*[21]	PSOC scale and satisfaction and efficacy subscales	A 16-item scale with two subscales. Parents respond to a series of questions about parenting, indicating their level of agreement or disagreement on a six-point Likert scale. The satisfaction subscale measures the extent that parents are satisfied with their role as a parent and so captures the affective dimension of parenting competence, including the extent of parental frustration, anxiety and motivation. The efficacy subscale measures the extent that parents feel that they are managing the role of being a parent and captures competence, problem-solving ability and capability in the parenting role. The authors provide a reference
Quine and Wade[146]	Improvement in knowledge of behavioural principles using Knowledge of Behavioural Principles as Applied to Children test	A 50-item multiple forced-choice test designed to assess understanding of the application of basic behavioural principles as they are applied to children, which takes about 30 minutes. Each item presents a problem situation to which the respondent is required to select the correct behavioural response. Criterion response for each question was selected on the basis of learning principles. The authors provide a reference

a As defined by study authors.

Appendix 14 Child-related outcomes for studies evaluating parent-directed non-tailored interventions

TABLE 41 Child-related outcomes for studies evaluating parent-directed non-tailored interventions

Study	Child sleep-related outcome assessed	Method of assessment	Definitions[a]
Global measures and composite scores			
Adkins *et al.*[127]	Sleep efficiency	Actigraphy	Percentage of TST/time in bed
	TST	Actigraphy	Actual time slept – the sum of all 'sleep epochs' measured in minutes within the interval between the time set on the actogram for night-time sleep and morning wake time
Beresford *et al.*[21]	CSHQ and subscales	CSHQ	The CSHQ was used to assess the severity of sleep problems in children aged 4–10 years. Parent reported. Subscales include bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night wakings, parasomnias, sleep disordered breathing and daytime sleepiness. The authors provide a reference
	Parent-set child sleep goals	Parent-set child sleep goals	A 10-point scale captured progress towards goals. Parents identified up to three goals during session 1 (GAS). The authors provide a reference
Beresford *et al.*[21]	CSHQ	CSHQ	The CSHQ was used to assess the severity of sleep problems in children aged 4–10 years. Parent reported. Subscales include bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night wakings, parasomnias, sleep disordered breathing and daytime sleepiness. The authors provide a reference
	Parent-set child sleep goals	Parent-set child sleep goals	A 10-point scale captured progress towards goals. Parents identified up to three goals during session 1 (GAS). The authors provide a reference
Malow *et al.*[128]	CSHQ	CSHQ	The CSHQ is a parent-completed questionnaire consisting of 33 questions on a 3-point scale. The CSHQ Is used to examine sleep behaviour in toddlers, preschool and school-aged children with a variety of conditions. Subscales of the CSHQ measure insomnia-related dimensions, such as bedtime resistance, sleep anxiety, sleep onset delay, sleep duration and night wakings. Other dimensions include daytime sleepiness, sleep disordered breathing and parasomnias. The authors provide a reference
	FISH	FISH	FISH is a quantitative scale of sleep habits, including bedtime routine, sleep environment and parental interactions. FISH as used was a 12-item scale – the full version includes 22 items. The authors provide a reference
	Sleep efficiency	Actigraphy and sleep diary	Per cent of TST out of the total time in bed

continued

TABLE 41 Child-related outcomes for studies evaluating parent-directed non-tailored interventions (*continued*)

Study	Child sleep-related outcome assessed	Method of assessment	Definitions[a]
	TST	Actigraphy	Actual time slept, the sum of all sleep epochs, measured in minutes with the interval between the time set on the actogram for night-time sleep and morning wake time
Montgomery *et al.*[49]	Composite Sleep Disturbance Score	Sleep diary	A Composite Sleep Disturbance Score was calculated by summing the score on each problem. Calculated from the sleep diary as follows: 4 = minimum entry score, representing, for example, a child with settling problems lasting > 30 minutes at least five times weekly and 8 = maximum score, representing a child who also wakes in the night for at least 30 minutes > 3 nights each week. No reference provided
Reed *et al.*[129]	CSHQ	CSHQ and subscales	The CSHQ is a parent-completed questionnaire used to examine sleep behaviour in toddlers, preschool and school-aged children with a variety of conditions. Subscales measured insomnia-related dimensions including bedtime resistance, sleep anxiety, sleep onset delay, sleep duration and night wakings. Additional dimensions include daytime sleepiness, sleep disordered breathing and parasomnias. A total score is also calculated and the authors reference as previously reported the use of a modified totals core incorporating the insomnia domains – total of items compromising the bedtime resistance, sleep anxiety, sleep onset delay, sleep duration and night wakings scales. The CSHQ was used to measure changes in subscales and total scores after the behavioural intervention. The authors provide a reference
	FISH	FISH	FISH is a parent-reported questionnaire that assesses sleep hygiene for their children. It includes giving attention to developing a structured consistent bedtime routine, sleep environment, daytime habits and parental interactions at bedtime and on night wakings. Parents rate the frequency of sleep habits over the last month on a 5-point scale. The authors previously validated a 12-item research version of the scale. In parent education classes, the authors used the 22-item scale that provides a comprehensive overview of sleep habits. The full version of the scale is provided in a table
	TST (time in bed)	Actigraphy	No definition provided
	Sleep efficiency	Actigraphy	No definition provided
Yu *et al.*[151]	CSHQ	CSHQ	The CHSQ is a sleep-screening instrument that is widely used to identify sleep problems in children with a range of problems, including ASD. It includes 50 questions, and each item is scored as 3 = usually (5–7 times/week), 2 = sometimes (2–4 times/week) or 1 = rarely (0–1 times/week). It produces a total score and eight subscale scores for sleep onset delay, night awakening, sleep duration, sleep resistance, sleep anxiety, parasomnia, daytime sleepiness and sleep disordered breathing, which reflect key sleep domains that encompass the major medical and behavioural sleep disorders. The authors provide a reference
	FISH	FISH	FISH was developed to measure sleep hygiene and behaviours in children with ASD. There are 22 questions and each question is scored from 1 to 5 depending on the frequency of occurrence, namely never, rarely, sometimes, usually and always. The authors provide a reference

TABLE 41 Child-related outcomes for studies evaluating parent-directed non-tailored interventions (*continued*)

Study	Child sleep-related outcome assessed	Method of assessment	Definitions[a]
Sleep initiation			
Adkins *et al.*[127]	Bedtime settling: SOL	Actigraphy	The number of minutes that it took for the child to fall asleep when the parent turned the lights out and expected them to fall asleep
Bramble[149]	Bedtime settling: time to settle (SOL)	Sleep diary	Mean time to settle once the child was in bed
Malow *et al.*[128]	Bedtime settling: SOL	Actigraphy and sleep diary	The number of minutes taken for the child to fall asleep when the parent turned the lights out and expected the child to fall asleep
Reed *et al.*[129]	Bedtime settling:		
	SL	Actigraphy	No definition provided
	Bedtime	Sleep diary and event markers	No definition provided
Sleep maintenance			
Adkins *et al.*[127]	Night waking:		
	WASO	Actigraphy	The total time that the child was awake during the night, after the SOL was excluded. WASO was measured as the sum of all wake epochs during the sleep period. WASO did not include wake time in bed before the final arising and terminal wakefulness was not encountered
	Fragmentation index	Actigraphy	The Fragmentation Index captures all movement regardless of the intensity of the movement. The Fragmentation Index is a measure of nocturnal movement that is calculated using: (number of movile epochs lasting four epochs + number of immobile epochs < 1-minute duration/number of immobile epochs > 1-minute duration) × 100
Bramble[149]	Non-specified night-time sleep disturbance: severity of sleep problems	Parent/carer report on VAS	Efficacy of treatment assessed by parental reports of the severity of sleep problems using a VAS (0 = no problems to 10 = very severe problems, converted to %). The severity of sleep problems was also rated on a categorical scale (0 = usually sleeps and settles well to 9 = sleep is disturbed, settles late and wakes early most nights). These scales were derived from the two sleep-problem items of the Wing and Goulds Handicaps, Behaviours and Skills scale. The authors provide a reference
Malow *et al.*[128]	WASO	Actigraphy and sleep diary (to confirm accuracy)	The total time that the child was awake during the night after the SOL was excluded. WASO was measured as the total of all wake epochs during the sleep period
Reed *et al.*[129]	Night waking: WASO	Actigraphy, sleep diary and event markers	No definition provided

continued

TABLE 41 Child-related outcomes for studies evaluating parent-directed non-tailored interventions (*continued*)

Study	Child sleep-related outcome assessed	Method of assessment	Definitions[a]
Child-related quality of life, daytime behaviour and cognition			
Bramble[149]	Child daytime behaviour and cognition	BPI	The BPI was derived for specific use with children with severe learning disabilities and was scored from 0 to 64. The authors provide a reference
Malow *et al.*[128]	Child-related quality of life	PedsQL (total score)	The PedsQL is a 23-item instrument designed for children aged 2–18 years. PedsQL includes four domains of functioning: physical, emotional, social and school. Each domain has a subscore and a total score and a psychosocial health summary score. The authors provide a reference
	Child daytime behaviour and cognition	CBCL	The parent-completed CBCL consists of two modules, one for ages 1.5–5 years and one for ages 6–18 years. For analysis, the authors selected muscles that were common to both modules and showed improvements in previous interventional studies (e.g. scales including anxious/depressed, withdrawn and withdrawn/depressed, attention and DSM-oriented scales attention deficit hyperactivity). The authors provide a reference
	RBS-R	RBS-R	The parent-completed RBS-R consists of six subscales: stereotyped, self-injurious, compulsive, ritualistic, sameness and restricted behaviours and a total scale. The scale is validated in children. Subscales were selected for analysis that had shown improvements in prior interventional studies (e.g. stereotyped, compulsive and restricted behaviours). The authors provide a reference
Reed *et al.*[129]	Child daytime behaviour and cognition	PCQ	The PCQ is a validated parent-completed questionnaire used to assess the presence and severity of 13 developmental and behavioural concerns expressed by parents of children with ASD. Domains include those related to core symptoms of ASD (e.g. language delay and social interaction) and related symptoms (e.g. hyperactivity and compulsive behaviours). The authors provide a reference
	RBS-R	RBS-R	The RBS-R is an observer-completed questionnaire validated in adults with ASD. A total score and scores for the subscales of stereotyped, self-injurious, compulsive, ritualistic, sameness and restricted behaviour are calculated. The authors provide a reference
Yu *et al.*[151]	Child daytime behaviour and cognition	CBCL	The CBCL measured the daytime behaviour of each child and is a commonly used questionnaire to identify social/emotional and/or behavioural problems in children. It is also used to aid diagnosis and evaluate emotional and behavioural problems in children with ASD. The form includes 100 problem items – 99 closed-ended items and one open-ended item that requests respondents add any additional problems not previously listed. Parents rate each item as 0 not true, 1 for somewhat true and 2 for very true or often true. A total problem score is calculated by totalling the scores on all of the items, including emotional reaction, anxious/depressed syndrome, somatic complaints, withdrawal, sleep problems, attention problems and aggressive behaviour. The internalising score is the sum of the scores on items in the withdrawal, somatic complaints and anxious/depressed syndrome profiles. The externalising score is the sum of the scores on the items on attention problems and aggressive symptoms. The authors provide a reference

TABLE 41 Child-related outcomes for studies evaluating parent-directed non-tailored interventions (*continued*)

Study	Child sleep-related outcome assessed	Method of assessment	Definitions[a]
	Daytime behaviour	PCQ	The PCQ is used to assess daytime behaviour. It is a 13-item parent-interview screening instrument assessing the severity of core developmental and associated psychiatric symptomology using a 4- point scale. Parents are asked to describe the extent that each symptom has been a problem with 1, 2, 3 and 4 representing no, mild, moderate and severe problems, respectively. The authors provide a reference

DSM, *Diagnostic and Statistical Manual of Mental Disorders*; GAS, Goal Attainment Score; VAS, visual analogue scale.
a As defined by study authors.

Appendix 15 Parent sleep-related outcomes for studies evaluating parent-directed non-tailored interventions

TABLE 42 Parent sleep-related outcomes for studies evaluating parent-directed non-tailored interventions

Study	Parent sleep-related outcome assessed	Method of assessment	Definitions[a]
Parent-carer-related quality of life			
Bramble[149]	Maternal stress	Rutters' Malaise Inventory	Rutters' Malaise Inventory measured changes in maternal stress and is scored from 0–11. The authors provide a reference
Reed *et al.*[129]	Parental stress	Parenting Stress Index – Short Form	A 36-item abbreviated version of the Parenting Stress Index, which provides a Total Stress Score and subdomain scores of parental distress, difficult child and parent–child interactions. The measure has been used to measure parental stress in autism disorder. The authors provide a reference
Yu *et al.*[151]	Parental stress	Parental Stress Index – Short Form	Used to assess parental stress. A validated reliable and widely used instrument for measuring parenting stress. Parents rate each of the 36 items on a 5-point scale ranging from strongly disagree to strongly agree. A total score is calculated. The authors provide a reference
Quality of sleep			
Bramble[149]	Maternal sleep quality	Maternal Sleep Scale	Mothers appraised their own sleep quality using an adapted version of De Diana's[175] sleep rating scale – the maternal sleep scale, which requires yes/no responses to 11 statements (e.g. 'I usually sleep well during the night') and is scored from 0 to 11. The authors provide a reference
Yu *et al.*[151]	Sleep quality	Pittsburgh Sleep Quality Index	The Pittsburgh Sleep Quality Index is used to assess parental sleep habit, quality and quantity in a range of populations. It is a self-rated questionnaire consisting of 19 questions that generate a total score and seven subscores, including sleep quality, SOL, sleep duration, habitual sleep efficiency, sleep disturbances, use of sleep medication and daytime dysfunction. The authors provide a reference

a As defined by study authors.

Appendix 16 Measures of perceived confidence and/or efficacy and/or understanding of sleep/sleep management for studies evaluating parent-directed non-tailored interventions

TABLE 43 Measures of perceived confidence and/or efficacy and/or understanding of sleep/sleep management for studies evaluating parent-directed non-tailored interventions

Study	Method of assessment	Definitions[a]
Beresford *et al.*[162]	PSOC (satisfaction and efficacy subscales)	The PSOC is a validated self-reported 17-item scale developed to assess parents' self-esteem. Two subscales provide a measure of self-efficacy, indicative of the parent's sense of his/her own problem-solving ability and capability as a parent, and a measure of satisfaction with parenting that reflects frustration, anxiety and motivation with the parenting role. A higher score indicates a higher parenting sense of competency
Beresford *et al.*[163]	PSOC (satisfaction and efficacy subscales)	The PSOC is a validated self-reported 17-item scale developed to assess parents' self-esteem. Two subscales provide a measure of self-efficacy, indicative of the parent's sense of his/her own problem-solving ability and capability as a parent, and a measure of satisfaction with parenting that reflects frustration, anxiety and motivation with the parenting role. A higher score indicates a higher parenting sense of competency
Malow *et al.*[128]	PSOC (satisfaction and efficacy subscales)	The PSOC is a validated self-reported 17-item scale developed to assess parents' self-esteem. Two subscales provide a measure of self-efficacy, indicative of the parents' sense of his/her own problem-solving ability and capability as a parent, and a measure of satisfaction with parenting that reflects frustration, anxiety and motivation with the parenting role. The authors provide a reference

a As defined by study authors.

Appendix 17 Child-related outcomes for studies evaluating non-comprehensive parent-directed interventions

TABLE 44 Child-related outcomes for studies evaluating non-comprehensive parent-directed interventions

Study	Child sleep-related outcome assessed	Method of assessment	Definitions[a]
Global measures and composite scores			
Peppers *et al.*[155]	CSHQ	CSHQ total score	Parent completed measure used to assess sleep in children who met the inclusion criteria and agreed to participate. A sleep disturbance score of ≥ 42 indicated a paediatric sleep disorder. The authors provide a reference
Wiggs and Stores[152]	Composite sleep index (bedtime resistance, night waking, early waking and sleeping in places other than bed)	Composite sleep index of the modified version of the Simonds and Parraga Sleep Questionnaire	Calculated from parental questionnaire information obtained through a modified version of the Simonds and Parraga Sleep Questionnaire. Scores ranged from 0 to 12. Settling and night waking were scored in terms of frequency and duration, and early waking and sleeping in the parents' bed for frequency only. Frequency = problems occurring more than several times a week = 2. Duration was scored as following: settling problems lasting up to 1 hour = 1; > 1 hour = 2. Night wakings were scored as if lasting a few minutes = 1 and if they lasted longer = 2. The authors provide a reference
	TST	Actigraphy	Time from sleep onset to the time the child woke up. Referred to as sleep period
Sleep maintenance			
Wiggs and Stores[152]	Non-specified night-time sleep disturbance:		
	• Movement during sleep • Movement index	Actigraphy	Defined as the sum of the epoch scores divided by total number of epochs in the sleep period
		Actigraphy	Defined as the number of thirties epochs with a value > 0 (i.e. with movement), divided by the total number of thirties epochs in the sleep period × 100
	Night waking:		
	• Fragmentation index	Actigraphy	Defined as the number of discrete thirties epochs with a value of 0 (i.e. with no movement) divided by the total number of immobile phases of any duration × 100
Child-related quality of life, daytime behaviour and cognition			
Peppers *et al.*[155]	ADHD symptoms	Parent Vanderblit ADHD symptom checklist	A parent-/caregiver-completed screening tool to assess ADHD behaviour. No further definition or reference provided
Wiggs and Stores[152]	Child daytime behaviour and cognition	ABC parent and teacher versions reported	The ABC was used to assess the following types of challenging behaviour: self injury, aggression, screaming, temper tantrums, non-compliance and impulsivity. The authors provide a reference

a As defined by study authors.

Appendix 18 Child sleep-related outcomes for studies evaluating other non-pharmacological interventions

TABLE 45 Child sleep-related outcomes for studies evaluating other non-pharmacological interventions

Study	Child sleep-related outcome assessed	Method of assessment	Definitions[a]
Global measures and composite scores			
Francis and Dempster[156]	TST	Sleep diary	No definition provided
Gringras *et al.*[36]	CSHQ	CSHQ (no data reported)	–
	Composite sleep disturbance index; frequency and duration of sleep problems	Composite sleep disturbance index; frequency and duration of sleep problems	No definition provided, reference to key papers provided
	Sleep efficiency	Parental diary	The proportion of time spent in bed asleep
	TST	Parental diary, actigraphy	No definition provided
Guilleminault *et al.*[158]	TST	Parent report (actigraphy verified)	Nocturnal TST
Oriel *et al.*[159]	CSHQ	CSHQ	No reference provided. The authors state that the CSHQ developed by researchers at Brown University was used to quantify sleep problems and consisted of eight subscales (bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night wakings, parasomnias, sleep disordered breathing and daytime sleepiness) and 33 items for the total sleep disturbance score. Maximum score of 99
	TST	Telephone call from researcher	The average number of hours of sleep for each participant during each study phase
Yu and Hong[161]	CSHQ	CSHQ	No reference provided, the authors state the scale was used to assess the sleep state of patients before and after treatment. Scale included: the total time of sleep' the time of getting into bed for sleep; sleep habit; sleep behaviour; wake state at night; state at getting up in the morning; sleep state at daytime and total score
Sleep initiation			
Francis and Dempster[156]	Bedtime settling: SL	Sleep diary	Time taken to fall asleep
Gringas *et al.*[36]	Bedtime settling: SOL	Parental diary, actigraphy	No definition provided

continued

TABLE 45 Child sleep-related outcomes for studies evaluating other non-pharmacological interventions (*continued*)

Study	Child sleep-related outcome assessed	Method of assessment	Definitions[a]
Oriel *et al.*[159]	Bedtime settling: SOL	Telephone call from researcher	The average number of minutes to fall asleep for each participant during each study phase
Piazza *et al.*[157]	Bedtime settling: exactly what time the child fell asleep at night	Observations	No definition provided
Sleep maintenance			
Francis and Dempster[156]	Night waking: time spent awake during the night	Sleep diary	'Where nightime awakenings for the child fell in the parent's sleep period, only those which disturbed the parent to awakening were recorded'[156]
Gringas *et al.*[36]	Night waking: number of night wakings		
	Time awake after sleep onset	Actigraphy and parental diary	No definition provided
	Proportion of nights with > 1 wakes	Actigraphy and parental diary	No definition provided
		Actigraphy and parental diary	No definition provided
Guilleminault *et al.*[158]	Non-specified night-time sleep disturbance: longest wake and sleep periods during 24-hour cycle	Sleep diary (actigraphy-validated sleep diary)	No definition provided
		Sleep diary (actigraphy-validated sleep diary)	No definition provided
Oriel *et al.*[159]	Night waking: number of night wakenings	Telephone call from researcher	No definition provided
Piazza *et al.*[234]	Night waking: time of night wakes and return to sleep	Observations	No definition provided
	Non-specified night-time sleep disturbance: hours of disturbed sleep	Observations	No definition provided
	Waking time: exactly what time the child awakened in the morning	Observations	No definition provided
Sleep scheduling			
Francis and Dempster[156]	Sleep quality	Sleep diary	No definition provided
Gringas *et al.*[36]	Sleep improvement	Children and parent blanket scale	No definition provided
	Quality of sleep	Child smiley-face rating and parent blanket scale	No definition provided
Guilleminault *et al.*[158]	Distribution of sleep bouts during 24-hour cycle	Parent report, actigraphy (validated sleep diaries)	No definition provided
Yehuda *et al.*[160]	Degree of fatigue in general during the day	Parent blanket scale	No definition provided
	Quality of sleep	Short questionnaire	No definition provided

TABLE 45 Child sleep-related outcomes for studies evaluating other non-pharmacological interventions (*continued*)

Study	Child sleep-related outcome assessed	Method of assessment	Definitions[a]
Child-related quality of life, daytime behaviour and cognition			
Francis and Dempster[156]	Child daytime behaviour and cognition	Sleep diary	No definition provided
Gringas *et al.*[36]	Daytime behaviour and cognition	ABC (total score and subscales)	No definitions provided, reference to checklist manual provided
	Sensory Behaviour Questionnaire	Sensory Behaviour Questionnaire; sensory stimuli response profile	Reference to unpublished original paper provided
Yehuda *et al.*[160]	Level of good mood in general	Short questionnaire	'5 point Likert scale'
	Level of ability to concentrate during the day, mainly at school		
	Percentage of homework completed in general		

SL, sleep latency.
a As defined by study authors.

Appendix 19 Studies evaluating family experience

TABLE 46 Studies evaluating family experience

Study	Sample	Experiences of receiving or implementing the intervention (parent report unless specified)	Parents' views on the value/relevance and/or usefulness of different elements of the intervention	Recommend the intervention?
Pharmacological interventions				
Wright *et al.*[106]	Sample: 20 study participants Data were recorded within usual trial data collection processes	One parent (1/20) withdrew their child from trial because they found that it was too difficult to administer the medication (oral melatonin)		Not specifically asked
Non-pharmacological: parent-directed tailored interventions				
Bramble[149]	Sample: 15/15 study participants Data collected: 4-month follow-up Fixed response question regarding style of intervention approach. Response options: too tough; rather tough; just right; rather soft; too soft Overall rating of helpfulness: visual analogue scale [0 (no help) to 10 (extremely helpful)] Checklist of 10 key components of intervention (referred to as 'advice items'): respondents ticked those that they felt had changed the child's sleep problems and rated helpfulness [+ 2 (very helpful) to –2 (very unhelpful)]	12/15 reported the treatment was 'just right'; 3/15 rated it 'rather tough' but were willing to continue	Overall rating of helpfulness: mean score 8.9 (SD 1.9) Mean ratings of helpfulness of 'advice items': • Parents back one another up, 1.64 • Pre-bedtime wind down, 1.40 • Setting and sticking to a regular bedtime, 1.34 • Ignoring child once in bed unless unwell, 1.27 • Regular settling routine, 1.13 • Bedroom made safe and secure, 1.07 • No-fuss prompt put-back of child during night, 1.00 • Bedroom light off and door closed, 0.93 • Short settling time and settling phase, 0.86 • Removing sources of stimulation from bedroom, 0.67	Not specifically asked
Austin *et al.*[123]	Sample: 5/6 study participants[a] completed 23-item CATS designed for study Response format: five-point Likert Scale (maximum score 35) Domains: treatment goals, training and resources, appropriateness and acceptability of intervention plan, treatment compliance, outcomes and implications, most valued aspects of programme, programme difficulties and suggestions for future sleep interventions	Domains of CATS: • ease of implementation • incorporation of parent priorities in plan development • stressfulness of the intervention plan Mean score 27.6 (SD 4.73) of a maximum score of 35 Three out of five parents reported that using bedtime restriction and/or bedtime fading with response cost was stressful to implement	Domains of CATS: • suitability, acceptability and relevance – mean score 31.6 (SD 1.82) • preparation, assistance and ability to understand training and resources – mean score 18.0 (SD 1.22). • improvement outcomes and usefulness of programme to address the problem – mean score 22.4 (SD 1.52)	5/5 would recommend

Study	Sample	Experiences of receiving or implementing the intervention (parent report unless specified)	Parents' views on the value/relevance and/or usefulness of different elements of the intervention	Recommend the intervention?

Johnson *et al.*[107]	Sample: 13/15 (active arm) and 17/18 (attention control)
	
	Parent Satisfaction Questionnaire developed for study. Parents rated the quality of various elements of the intervention:
	
	• number and length of sessions
	• usefulness of teaching tools (e.g. video vignettes, in-session worksheets and homework)
	• the helpfulness of specific elements
	
	Response format: three- or four-point Likert scales (higher scores reflecting greater levels of satisfaction)
	
	Adherence to intervention:
	
	• clinician-completed checklists of observations of evidence of completion of homework and implementation of recommended strategies
	• attendance rates at intervention programme recorded
		Parent Satisfaction Questionnaire
		
		• Overall satisfaction rating:
		  ○ Active arm – 90% (range 70–100%)
		  ○ Attention control – 88.2% (range 63–100%)
		
		Clinician report
		
		• Adherence:
		  ○ Active arm – 93% (range 75–100%)
		  ○ Attention control – 98% (range 75–100%)
		
		• Attendance:
		  ○ Active arm – 73/75 sessions attended by all parents (97.3%)
		  ○ Attention control – 89/90 sessions attended by all parents (98.9%)
			Parents in the active arm only
			
			Helpfulness of specific elements of the intervention:
			
			• 69% reported that behaviour principles training was very helpful
			• 89% reported that the session on 'addressing prevention techniques and bedtime routines'[107] was very helpful
			• 77% reported that the session on 'addressing reinforcement and extension procedures for bedtime struggles, night awakenings and early morning awakenings'[107] was very helpful
			• 77% reported that the session on 'addressing sleep onset and sleep association procedures'[107] was very helpful
			• 61% reported that the 'booster and maintenance session'[107] was very helpful
			
			All other responses were 'okay'; there were no ratings of 'not helpful'
				Not specifically asked

continued

TABLE 46 Studies evaluating family experience (*continued*)

Study	Sample	Experiences of receiving or implementing the intervention (parent report unless specified)	Parents' views on the value/relevance and/or usefulness of different elements of the intervention	Recommend the intervention?

Moss *et al.*[124]	Sample: 26/26 study participants
	
	Completed 'an informal survey designed by study authors assessing acceptability of the programme's goals, workshops and resources, treatment plans, and outcomes'.[124] Referred to as CATS. Response format: five-point Likert scale. Total number of items not reported. Not clear if same as used by Austin *et al.*[123]
	
	Subsample (6/26) participated in a semistructured interview. (20/26 not available for interview.) Method of data analysis not reported
		Parent report
		
		• Responses to CATS:
		  ○ satisfaction with intervention – 21/26 responded satisfied or very satisfied
		  ○ programme goals – 24/26 responded acceptable or very acceptable
		  ○ implementation of the treatment plans – 21/26 responded acceptable or very acceptable
		  ○ workshop materials – 24/26 responded acceptable or very acceptable
		  ○ programme worthwhile – 24/26 responded mostly worthwhile or completely worthwhile
		
		Semistructured interviews
		
		• Elements of the programme that were spontaneously mentioned as what was 'least liked' about the intervention:
		  ○ nothing to report – 5/6
		  ○ early morning time of workshop – 1/6
		
		• None felt that there was any aspect of the intervention that needed to be improved
		• Perceived barriers to other parents completing the intervention:
		  ○ time pressures or putting in initial time commitment needed for the intervention – 5/6
		  ○ reluctance or fear of change – 2/6
			Semistructured interviews
			
			• Elements of programme that were spontaneously mentioned as what was 'most liked' about the intervention:
			  ○ follow-up support – 5/6
			  ○ workshops – 2/6
			  ○ individualised approach – 2/6
			
			• Elements of the child's sleep management plan that were spontaneously identified as 'most useful':
			  ○ communication strategies – 5/6
			  ○ sensory strategies – 2/6
			  ○ behavioural strategies – 2/6
				Not specifically asked

Study	Sample	Experiences of receiving or implementing the intervention (parent report unless specified)	Parents' views on the value/relevance and/or usefulness of different elements of the intervention	Recommend the intervention?
Scibberas *et al.*[125]	Sample: 27/27 study participants asked to complete a study-designed scale. The number completing the scale not reported The 'study-designed scale' captured parents' reports of the helpfulness of the intervention and use sleep management strategies learnt during the intervention. No further information provided	'Most caregivers reported that they could implement sleep management strategies "at least half of the time" '[125] (p. 934). Note: 'most' not further specified	'Most . . . reported sleep strategies as helpful, including: • advice about normal sleep (94%) • setting bedtimes (94%) • sleep hygiene (87%) • using a sleep diary (93%) • a sleep plan (82%) • limit setting (93%), using rewards (85%) • relaxation strategies (83%) • returning the child to bed overnight (75%) • checking method (73%) • bedtime fading (67%)'[125] (p. 934)	'All but one'[125] (p. 934) would recommend the intervention

Weiskop *et al.*[126]	Sample: 12/12 study participants
	
	Parents completed a modified version of the PEQ (Griffin and Hudson, 1978[235]), which comprised:
	
	• Three open-ended questions about the programme:
	  ○ what the parents liked best?
	  ○ what the parent liked least?
	  ○ what they would change?
	
	• Five items (5-point Likert scale response):
	  ○ approval of techniques[b]
	  ○ improvements seen in child's sleep[b]
	  ○ improvements in child's general behaviour
	  ○ reduction in parental stress levels
	  ○ how strongly parent would recommend intervention programme
	
	• Approval of techniques[b]
		Approval of techniques: 7/12 gave the maximum positive rating
		
		Overall satisfaction: mean score 13.8 (range 11–15)
		
		Least-liked elements were:
		
		• 'sticking to a bedtime routine'[126] – 2/12
		• the sessions were too long – 1/12
		• referred to the 'overall time-consuming nature of the programme'[126] (p. 102) – 3/12
			'Responses to the PEQ indicated that the best aspects of the programme were . . . the support provided, the telephone calls, and the method of instruction'[126] (p. 102)	12/12 would recommend

continued

TABLE 46 Studies evaluating family experience (*continued*)

Study	Sample	Experiences of receiving or implementing the intervention (parent report unless specified)	Parents' views on the value/relevance and/or usefulness of different elements of the intervention	Recommend the intervention?
Non-pharmacological: parent-directed non-tailored interventions				
Adkins *et al.*[127]	Sample: 6/9 study participants in active arm ($n = 9$) Parents in the active arm (who received a sleep education pamphlet) were 'asked a series of questions to collect parent feedback on pamphlet use'[127] (p. S141) and what was 'most useful about the pamphlet and what might have been more useful'[127] (p. S140)	Parents commented that the pamphlet contained good information, but that it would have been more useful to be given specific examples of how to take the information and put it into practice	Feedback from parents suggested that the pamphlet was useful as it contained good information, for example they liked that it included 'basic rules for sleep' and 'important of consistent bedtime'. They commented that they would have found it more useful if it had included specific suggestions of how to take the information and put it into practice	Not specifically asked
Malow *et al.*[128]	Sample: 80 study participants. (individual-mode arm, $n = 41$; group-mode arm, $n = 39$) Parents completed an 'anonymous survey' at end of education element of intervention. A 4-point Likert scale was used to capture views on general satisfaction with content of intervention and educator, whether or not they would recommend it to others, whether or not the intervention had improved the child's sleep habits and whether or not they would have preferred alternative mode of intervention delivery	Preferences for alternative mode of delivery: • Individual-mode arm – 3/41 reported would have preferred group delivery • Group-mode arm – 8/39 reported would have preferred a one-to-one session	Response to statement: 'The information covered was relevant and useful' • Individual-mode arm – 35/41 • Group-mode arm – 29/39 Authors report: 'There were no differences between responses in the "end of evaluation" survey based on mode of parent education'[128] (p. 223)	'Strongly agreed' that they would recommend: • individual-mode arm – 38/41 • group-mode arm – 32/39
Reed *et al.*[129]	Sample: 18/20 study participants Parents completed an 'anonymous evaluation'. No further details were given	Duration of the workshop was sufficient: 10/18	The information conveyed was relevant and useful: 17/18	18/18 would recommend
Montgomery *et al.*[49]	Sample: 23/33 study participants who received copy of booklet regarding managing sleep Brief questionnaire was given to evaluate the booklet in terms of relevance, ease with which it can be understood and usefulness on a four-point Likert scale (low = poor/negative experience; high = good/positive experience). The maximum score was 12		Mean total score on the questionnaire evaluating the booklet: 10.17 (SD 1.87)	Not specifically asked

Study	Sample	Experiences of receiving or implementing the intervention (parent report unless specified)	Parents' views on the value/relevance and/or usefulness of different elements of the intervention	Recommend the intervention?
Non-pharmacological: parent-directed: two tailored and two non-tailored				
Beresford *et al.*[130]	Sample: 35 parents purposively sampled (in terms of intervention outcome, child's diagnosis, parents' education and partner involvement in the intervention) from a total sample of parents ($n = 74$) who had received one of four parent-directed interventions included in this review: • two tailored interventions ($n = 12$); • two non-tailored [a half-day workshop ($n = 8$) and a four-session (one per week), group-delivered intervention ($n = 15$)] Semistructured interviews (individual telephone interviews and focus groups) were used to gather data on parents' views and experiences in terms of (1) parents' descriptions of the processes by which a sleep management intervention leads to improvements in their child's sleep, (2) parents' views of the factors that hinder the achievement of positive intervention outcomes and (3) parents' views on intervention intensity and mode of delivery	Parents referred to the demands that the intervention places on parent/caregivers, including the challenge of changing and sustaining new sleep management strategies Implementing new approaches to managing their child's sleep could be hampered by a number of factors: (1) a lack of consistency across caregivers, (2) changes and disruptions in usual routines (e.g. illness, holidays) and (3) issues with the home environment, particularly when the child shared a bedroom siblings)	Features of the intervention identified by parents as supporting positive outcomes: • information that changed or developed parents' understanding of sleep and its management, and clarifying any links between the child's condition and sleep issues • attention being paid to building and developing parents' sense of competence and ability to make changes to the way they managed their child's sleep • learning that many parents of disabled children experience sleep management issues (this was particularly pertinent for parents receiving group-delivered interventions) • training on sleep management behaviours and strategies. Two key areas of change described: setting up or improving bedtime routine and handling night wakings • parents who were receiving sleep management interventions delivered over a number of weeks spoke about the value of ongoing support to help them persevere with new or changed routines • several parents said that keeping a sleep diary kept them motivated by revealing progress over time • improvements in their own well-being through getting better sleep themselves, this allowed them to have more energy and mental resources to further implement changes in sleep management	Not specifically asked

continued

TABLE 46 Studies evaluating family experience (*continued*)

Study	Sample	Experiences of receiving or implementing the intervention (parent report unless specified)	Parents' views on the value/relevance and/or usefulness of different elements of the intervention	Recommend the intervention?
			Features of the intervention identified by parents as hindering positive outcomes: • training not sufficiently tailored to child's particular needs and abilities. (Reported by those receiving non-tailored interventions, particularly a single workshop)	
Other non-pharmacological interventions				

Gringras *et al.*[36]	Sample: 73 study participants. Data recorded within usual trial data collection processes	Child report
		• Responses to the 'Children's Blanket Scale': Which best describes how you have felt about the blue blanket you have been using over the past 2 weeks?
		  ○ really liked the blanket: 48 vs. 31% (weighted blanket vs. control)
		  ○ blanket was just OK: 37 vs. 39% (weighted blanket vs. control)
		  ○ really disliked the blanket: 15 vs. 29% (weighted blanket vs. control) ($p = 0.110$)
			Parents' views on the value/relevance and/or usefulness of different elements of the intervention: parents favoured the weighted blanket	Not specifically asked

CATS, Caregivers Acceptance of Treatment Survey; PEQ, Program Evaluation Questionnaire.
a In controlled study, this refers to the number of participants in the active arm.
b Scores combined to give an overall measure of participant satisfaction (maximum score 15).

Appendix 20 Study quality: studies of acceptability/feasibility and experiences of implementing sleep interventions

TABLE 47 Study quality: studies of acceptability/feasibility and experiences of implementing sleep interventions

Study	Abstract and title	Introduction and aims	Method and data	Sampling	Data analysis	Ethics and bias	Results	Transferability/ generalisability	Implications and usefulness	Score (maximum 36)
Beresford *et al.* (2016)[130]	Good	Good	Good	Fair	Good	Fair	Good	Good	Good	34
Comments: sample drawn from sample recruited to outcomes evaluations. The recruitment to the outcomes evaluations had limitations										
Bramble (1996)[122]	Poor	Fair	Fair	Poor	Poor	Poor	Fair	Fair	Fair	23
Comments: lacks clear description of presentation of questions and method of recording. No report of parent characteristics. No account given of data analysis. Ethics approval not reported. Some further analyses needed/reported, for example table 3. Evaluation of a specific intervention. Findings not (necessarily) transferable to other parent-directed interventions. No mention of authors' biases – study conducted by one author with clinical experience										

Appendix 21 Study quality: quality assessment of non-pharmacological interventions

Summary of quality assessment using the Cochrane risk of bias for randomised controlled trials tool[62]

TABLE 48 Summary of quality assessment using the Cochrane risk of bias tool for RCTs

Study	Domain						Additional questions for crossover trials
	1. adequate sequence generation?	2. allocation concealment?	3. blinding?	4. incomplete outcome data addressed?	5. free of selective reporting?	6. free of other bias?	
Parent-directed tailored interventions							
Beresford *et al.*[21]	Unclear No details are provided	Unclear No details are provided	No No details are provided, but it would have been difficult to achieve blinding given the nature of the intervention	No Response rates were reported as 92% (post intervention) and 62% (12-week follow-up) (i.e. 5/12 lost to follow-up). No reasons for loss to follow-up were given or if/how this was dealt with in the analysis. No indication was given of which arm of the trial the participants were lost from	Unclear No protocol but data analysis detailed in appendix E. It seems to be an explanation of what analyses were carried out rather than an a priori plan/protocol. This intervention is part of larger study of multiple interventions – the same analyses will be carried out on all interventions, as applicable. All measures appear to have been reported	No Small study sample ($n = 13$). The intervention was suspended on two occasions, which affected the small sample size	N/A
Hiscock *et al.*[138]	Yes Computer-generated random number sequence	Yes Sealed opaque envelopes	No Parents were aware of the intervention (see discussion in the paper). It would be difficult or impossible to blind given nature of intervention	Yes Missing data were imputed, and both imputed and non-imputed reported Intervention and control group at baseline: $n = 122$ in each group Number lost to follow-up at the 3-month follow-up: intervention, $n = 36$; control, $n = 33$ Number lost to follow-up at 6-month follow-up: intervention, $n = 16$; control, $n = 33$	Yes Protocol available. All specified outcomes reported	Yes	N/A

Study	Domain						Additional questions for crossover trials
	1. adequate sequence generation?	2. allocation concealment?	3. blinding?	4. incomplete outcome data addressed?	5. free of selective reporting?	6. free of other bias?	
				Numbers included in the primary analyses varied for parent-and teacher-reported ADHD symptoms ($n = 99$ and $n = 83$ in the intervention group; $n = 85$ and $n = 77$ in the control group, respectively) Multiple imputation used in intention-to-treat analysis. There were $n = 122$ in each group			
Johnson *et al.*[107]	Unclear The authors state that participants were equally randomised using block randomisation with a block size of 10, but no information is provided about how they generated a sequence	Unclear No details of this are provided	No Parents and therapists were not blinded	No Only participants for whom baseline and 4-week follow-up data were available were included in the analysis	Unclear No protocol was referenced in the paper and one was not found after searching. The outcomes reported are those that are expected. However, without a protocol, it is difficult to be certain	Yes	N/A
Moss *et al.*[124]	Unclear No details are provided	Unclear No details are provided. Allocation was by research team but it is not clear if any attempts were made to conceal this (e.g. opaque envelopes)	No Owing to the nature of the intervention, participants and staff would know which group they were in	Unclear A total of 26 children were recruited: post treatment, $n = 22$ (treatment group, $n = 12$; control group, $n = 10$); follow-up, $n = 18$ (treatment group, $n = 10$, control, $n = 8$). Reasons for dropout (from the control and intervention groups after treatment) were family work/carer commitment, child health problems or family tragedy. There was no mention of pre-intervention dropouts	Unclear No protocol was referenced in the paper and one was not found after searching. The outcomes reported are those that are expected. However, without a protocol, it is difficult to be certain	Yes One child in sample was taking melatonin, but we do not think this would be enough to introduce bias	N/A

continued

TABLE 48 Summary of quality assessment using the Cochrane risk of bias tool for RCTs (*continued*)

Study	Domain						Additional questions for crossover trials
	1. adequate sequence generation?	2. allocation concealment?	3. blinding?	4. incomplete outcome data addressed?	5. free of selective reporting?	6. free of other bias?	
Sciberras *et al.*[125]	Yes	Unclear	No	Unclear	Unclear	Yes	N/A
	Computer-generated random number sequence	Allocation was done by independent statistician, but there is no detail on the process of this and if/how it was concealed	Owing to the nature of the intervention, the researchers and participants would know which group they were in	The authors do not state how loss to follow-up was addressed ($n = 8$ at 2 months and $n = 4$ at 5 months)	No protocol was referenced in the paper and one was not found after searching. The outcomes reported are those that are expected. However, without a protocol, it is difficult to be certain		
Parent-directed non-tailored interventions							
Adkins *et al.*[127]	Unclear	Unclear	No	Unclear	Unclear	Unclear	N/A
	No details are provided	No details are provided about allocation concealment	Blinding would have been difficult to achieve – both to parents and those taking the assessments/ collecting data. First, parents receiving the leaflet intervention would know that they were in that intervention, and those not receiving a leaflet would know they were in the control group. In the discussion, the authors state they could have used a generic leaflet for the control group to promote better blinding. In addition, the clinicians/ researchers knew who received the leaflet because they instructed parents to read it	A total of 18 were randomised into each group ($n = 36$), but in table 2, it says there are $n = 19$ in the pamphlet condition and $n = 17$ in the no pamphlet condition. There is no explanation in the text about this discrepancy and, therefore, it is not clear if the loss of one participant in one group means that there were missing data, and if and how these were dealt with	No protocol was referenced in the paper and one was not found after searching. The outcomes reported are those that are expected. However, without a protocol, it is difficult to be certain	The authors say that the leaflet used for the intervention is available from a website. Is it possible that the leaflet was available on this website pre intervention and, therefore, parents could have accessed it before the trial, which would introduce bias? It is impossible to clarify this, as it is not clear if the leaflet was already in use and available publicly	

Study	Domain						Additional questions for crossover trials
	1. adequate sequence generation?	2. allocation concealment?	3. blinding?	4. incomplete outcome data addressed?	5. free of selective reporting?	6. free of other bias?	
Malow *et al.*[128]	Yes Database software randomly assigned participants in a 1 : 1 block in each site	Unclear Allocation was carried out using software but still not clear in the paper if this was concealed from the researchers	No Participants were not blinded. It would have been difficult to do given nature of the intervention	No A total of 114 participants were enrolled into study. Of these, 80 completed all study procedures. Only the 80 with complete data were included in analysis. Reasons for non-completion were that it was too time-consuming, SOL not confirmed by actigraphy, children could not tolerate the actigraphy device, the child started new medications or parents opted for other ways of addressing sleep problems In total, 41 were randomised to the group arm and 39 to the individual arm. Six families switched from the group arm to the individual arm because of logistical reasons. Analysis was made with and without switchover	Unclear No protocol was referenced in the paper and one was not found after searching. The outcomes reported are those that are expected. However, without a protocol, it is difficult to be certain	Yes Six participants randomised to the individual arm were then moved to the group arm. This could introduce bias, but the authors analysed and present results both with and without these six participants in the individual arm. After reporting results from the two arms (both intervention groups, no control group), they combined data from the two and report together as before and after. However, this is not a bias in the design, but a possible bias in the presentation of the results	N/A

continued

TABLE 48 Summary of quality assessment using the Cochrane risk of bias tool for RCTs (*continued*)

Study	Domain						Additional questions for crossover trials
	1. adequate sequence generation?	2. allocation concealment?	3. blinding?	4. incomplete outcome data addressed?	5. free of selective reporting?	6. free of other bias?	
Montgomery *et al.*[49]	Unclear No details are provided	Unclear Opaque envelopes were used but there was no evidence that envelopes were sequentially numbered. Envelopes were 'selected' by an independent researcher	No There was no blinding of participants, which would have been difficult given nature of the intervention	Yes A total of 2/66 were missing at follow-up owing to families moving. Families moving is not related to the study treatment and so this is unlikely to affect fidelity of outcomes	No No protocol was referenced in the paper and one was not found after searching. The paper reports Composite Sleep Disturbance Score as the main outcome and evaluation of the programme, but we would expect to see objective measures of sleep	No No detail on age or gender of the two groups and, thus, comparability of groups. Not clear what type of statistical analysis was used in some places	Was use of a crossover design appropriate? Yes Is it clear that the order of receiving treatments was randomised? Yes Can it be assumed that the trial was not biased from carry-over effects? There would be no possibility of carry-over effect as the only crossover is from the control group to the treatment group Are unbiased data available? A Kruskal–Wallis test is used
Other parent-directed interventions							
Wiggs and Stores[152]	Unclear No details are provided, other than that the schools were randomised. The number of 'clusters unclear'	Unclear No details are provided	No Owing to the nature of the intervention, the researchers and participants would know which group they were in. The paper does not describe the study as blinded	Unclear No details are provided as to whether or not there was loss to follow-up	Unclear No protocol was referenced in the paper and one was not found after searching. The outcomes reported are those that are expected. However, without a protocol, it is difficult to be certain	No Reporting of the trial design was unclear. It was also unclear how the control group was 'matched', as the trial randomised schools (clusters) but matched intervention and control children (individuals) It is not clear if the analysis accounted for the study being a cluster trial. The authors state that there was no explicit hypotheses but data were analysed in an 'exploratory manner'	N/A

Other non-pharmacological interventions							
Piazza *et al.*[157]	Unclear No details are provided	Unclear No details are provided	No Owing to nature of the intervention, assessors would know which group (of the two treatment groups) participants were in. Data were collected in a sleep laboratory by trained observers and the potential for bias in recording was mitigated by having two observers on 86% of days across all clients	Unclear Data were presented for 14 participants. There were no details about the number of participants who were initially approached/ recruited or if it was ≥ 14	Unclear No protocol was referenced in the paper and one was not found after searching. The outcomes reported are those that are expected. However, without protocol, it is difficult to be certain	Yes	N/A
Francis and Dempster[156]	Yes Random numbers table	Unclear Allocation was done by a senior investigator. The paper reports that this senior investigator had no contact with the participating children and parents (who self-completed the data collection). Codes were known only to the senior investigator. But, it is not clear if this process was concealed to the investigator	Unclear Investigator who held the codes had no contact with participants and the placebo and treatment capsules were similar in appearance; therefore, blinding to participants is plausible. It is possible that research staff (except the senior investigator) were blinded, as only the senior investigator had the key to the codes, but it is also not clear what involvement this investigator had in the subsequent research process and whether or not this would introduce bias The authors state that there was blinding by the senior investigator who had no contact with patients or parents. Outcomes were self-reported so the outcome is unlikely to be influenced	Yes There was no loss to follow-up and a total of five participants	Unclear No protocol was referenced and one was not found after searching. The study is listed on European Union Clinical Trials Register, but little information is provided on the study records and there is not enough to make a judgement about whether or not all outcomes are reported. However, relevant sleep outcomes are reported The description of measures other than primary outcomes is poor. There is no detail of what was included in end-of-study interviews; therefore, it is difficult to determine whether or not there was selective reporting	No Only night wakings that occurred during parents sleep period were recorded; therefore, other night wakings outside the parent sleep period would not be recorded. Authors say these data must be regarded as conservative Some of the children were taking other medications for sleep The study had very small numbers ($n = 5$). There are no details of the recruitment process other than that it was through schools/ organisations for children with LD. There is no indication of the total numbers approached or if these five patients self-selected	Was use of a crossover design appropriate? Yes Is it clear that the order of receiving treatments was randomised? Yes Can it be assumed that the trial was not biased from carry-over effects? Yes, washout period used, but no analysis of carry-over effect Are unbiased data available? Yes. A repeated measures ANOVA was used

continued

TABLE 48 Summary of quality assessment using the Cochrane risk of bias tool for RCTs (*continued*)

Other non-pharmacological interventions							
Gringras *et al.*[36]	Unclear Block randomisation with random variable block lengths of 2 and 4, stratified by centre, but no detail of sequence generation was given	No Authors report that the trial investigators and the statistician were blind to treatment allocation, but researchers were not. Although randomisation was done remotely, the researchers dispensed the allocated treatments	No Researchers were not blinded to initial allocation but trial investigators and the statistician were all blinded throughout the trial and analysis It is impossible to blind parents/children to the weight of the blankets but all other aspects of the blankets were the same	No A total of $N = 73$ were randomised. The loss to follow-up $n = 0$. Discontinued intervention and not included in the analysis $n = 6$ The reasons were that the child could not tolerate the blanket ($n = 4$, not clear which arm of trial these participants were from), the child was ill ($n = 1$) or parent withdrew child ($n = 1$). There was no discussion of the effect on analyses of loss resulting from not being able to tolerate blanket but this is a key finding and so likely to have an impact on results A total of $n = 13$ were excluded from analysis owing to insufficient or missing data. Missing data for questionnaire were prorated if < 10% were missing, or excluded otherwise. There was no detail on the reasons for missing data	Unclear Tried to access the protocol (the URL to this is listed in the paper), but when clicking on the link it takes you to a web page about ongoing projects. From this unable to find the protocol. The study reports all the outcomes expected, but it is difficult to be certain without protocol	Yes	Was use of a crossover design appropriate? Yes Is it clear that the order of receiving treatments was randomised? Yes Can it be assumed that the trial was not biased from carry-over effects? The authors report (p. 299) that there were no breaks between interventions, and so no 'washout' period. Although this is not a study of a drug, there might have been a carryover of the effects of the weighted blanket. For example, if the child had grown accustomed to better sleep? Are unbiased data available? Data were presented as baseline, weighted and control, regardless of order. Authors use an independent *t*-test to rule out a period effect and then undertook a paired *t*-test

ANOVA, analysis of variance; LD, learning disability; N/A, not applicable.
Yes, low risk of bias; unclear, unclear risk of bias; no, high risk of bias.

Summary of quality assessment for controlled before-and-after studies using ACROBAT-NRSI[63]

TABLE 49 Summary of quality assessment for controlled before-and-after studies using ACROBAT-NRSI

Other non-pharmacological intervention (Yehuda *et al.*[160])			
Bias because of confounding			
Domain	**Reviewer 1 quality appraisal**	**Reviewer 2 quality appraisal**	**Reviewers 3 and 4 quality appraisals**
1.1 Is confounding of the effect of the intervention unlikely in this study?	PN A total of 40 children received treatment and 38 received placebo. There are no details on why participants were allocated (if they were allocated) to each group. The characteristics of participants is not provided in detail (only overall age range, diagnosis of ADHD and reported sleep deprivation). A non-ADHD group served as a control group. The group was reported to have corresponding ages and socioeconomic statuses but details were not provided	NI It is hard to judge given the little information about the sample. The sample were selected based on whether or not they had ADHD, were male and were sleep deprived. I cannot think of a prognostic factor (other than the above, which were the reasons for receiving the intervention) that would predict the participants receiving the sleep intervention (fatty acids). One possibility might be whether or not there was a nutritional intolerance to fatty acids (which might predict if someone would not get the intervention) but this is not reported	Guidance for ACROBAT-NRSI, states that NI is not an option for this question. I would agree with KB that PN is appropriate as there is some information, albeit a limited amount of it is provided. Agree with KB and AS that PN is appropriate given that very limited information is provided on allocation and participants
1.2. If N or PN to 1.1: Were participants analysed in accordance with their initial intervention group throughout follow-up?	Y	NI It appears so but there is no information about dropout or whether or not participants switched groups	NI – insufficient information provided to enable judgement
1.3. If N or PN to 1.2: Were intervention discontinuations or switches unlikely to be related to factors that are prognostic for the outcome?	NI – insufficient information provided to enable judgement	N/A	NI – insufficient information provided to enable judgement

continued

TABLE 49 Summary of quality assessment for controlled before-and-after studies using ACROBAT-NRSI (*continued*)

Other non-pharmacological intervention (Yehuda *et al.*[160])			
Bias because of confounding			
Domain	**Reviewer 1 quality appraisal**	**Reviewer 2 quality appraisal**	**Reviewers 3 and 4 quality appraisals**
If Y or PY to 1.3, answer questions 1.4 to 1.6, which relate to baseline confounding. If N or PN to 1.1 and 1.2 and 1.3, answer questions 1.7 and 1.8, which relate to time-varying confounding			
1.4. Did the authors use an appropriate analysis method that adjusted for all the critically important confounding domains?	NI	N/A	N/A – in line with guidance
1.5. If Y or PY to 1.4:	N/A	N/A	N/A – as per above guidance
Were confounding domains that were adjusted for measured validly and reliably by the variables available in this study?			N/A – in line with guidance
1.6. Did the authors avoid adjusting for post-intervention variables?	Y	N/A	N/A – as per above guidance
	Authors presented before and after scores for each group with no adjustments		N/A – in line with guidance
1.7. Did the authors use an appropriate analysis method that adjusted for all the critically important confounding domains and for time-varying confounding?	N/A	N/A	N/A
			N/A – in line with guidance
1.8. If Y or PY to 1.7:	N/A	N/A	N/A
Were confounding domains that were adjusted for measured validly and reliably by the variables available in this study?			N/A – in line with guidance

Other non-pharmacological intervention (Yehuda *et al.*[160])			
Bias because of confounding			
Domain	**Reviewer 1 quality appraisal**	**Reviewer 2 quality appraisal**	**Reviewers 3 and 4 quality appraisals**
Bias in selection of participants into the study			
2.1. Was selection into the study unrelated to intervention or unrelated to outcome?	N Children in the intervention and placebo groups were selected into study because of ADHD and sleep deprivation. In total, 6/7 outcomes related to ADHD and/or sleep deprivation The comparison group was unrelated to the intervention or the outcomes	NI There is not enough information in the paper about recruitment to make a judgement on this. This applies to all the outcomes	N – some but very limited information is provided explaining that children were recruited on the basis of ADHD and sleep deprivation
2.2. Do start of follow-up and start of intervention coincide for most subjects?	Y The intervention lasted 10 weeks. Questionnaires were completed on day 1 and at end of 10 weeks	Y Participants answered a questionnaire/data collected on day 1 of study. This was applicable for all outcomes, which were assessed by the same questionnaire	Y – participants completed a questionnaire on the first day of the study and at the end of the 10-week intervention period
2.3. If N or PN to 2.1 or 2.2: Were adjustment techniques used that are likely to correct for the presence of selection biases?	N/A Overall judgement = ?. Selection into the study was not related to intervention/ essential fatty acids but was related to outcome, but when testing the impact it has on sleep, you have to select those having trouble with sleep	N/A	PN? N/A

continued

TABLE 49 Summary of quality assessment for controlled before-and-after studies using ACROBAT-NRSI (*continued*)

Other non-pharmacological intervention (Yehuda *et al.*[160])			
Bias because of confounding			
Domain	**Reviewer 1 quality appraisal**	**Reviewer 2 quality appraisal**	**Reviewers 3 and 4 quality appraisals**
Bias in measurement of interventions			
3.1 Is the intervention status well defined?	Y Two capsules/day of an essential fatty acids mixture composed of alpha-linolenic 0.95 g/ml and linolenic 0.90 g/ml free fatty acids, both 99% pure. Each capsule contained 360 g of linolenic acid and 90 g of alpha-linolenic acid in mineral oil. The placebo was composed of mineral oil in an identical capsule	Y Detail of the treatment is given on p. 1167. It applies to all outcomes	Y – A specific mixture of essential fatty acids was used that were created in the researchers' laboratory. Highly purified alpha-linolenic and linoleic acids were used to avoid the variations that occur in commercially prepared fatty acid oils, which may introduce possible confounding effects of other fatty acids or lipid mixtures. The essential fatty acids mixture was composed of alpha-linolenic (0.95 g/ml) and linoleic (0.90 g/ml) free fatty acids, both 99% pure. Each capsule contained 360 g of linoleic acid and 90 g of alpha-linoleic acid in mineral oil. The placebo was mineral oil in an identical capsule. The treatment lasted 10 weeks and each participant took two capsules per day Y
3.2 Was information on intervention status recorded at the time of intervention?	Y Given that capsules were created to be identical in the intervention and placebo, participants must have been allocated to intervention or placebo group; therefore, the group was known at start of intervention	NI There is not enough detail in the paper to make a judgement about this	PY – Given that capsules were created to be identical in control and placebo, participants must have been allocated to the intervention or placebo group; therefore, group was known at start of intervention PY
3.3 Was information on intervention status unaffected by knowledge of the outcome or risk of the outcome?	Y This was an experimental study with groups assigned	NI There is not enough detail in the paper to make a judgement about this	NI – there is not enough detail in the paper to make a judgement about this NI – insufficient information

Other non-pharmacological intervention (Yehuda *et al.*[160])			
Bias because of confounding			
Domain	**Reviewer 1 quality appraisal**	**Reviewer 2 quality appraisal**	**Reviewers 3 and 4 quality appraisals**
Bias because of departures from intended interventions			
4.1. Were the critical cointerventions balanced across intervention groups?	NI No data were available on possible cointerventions, that is, nothing on what other drugs/behavioural methods were being used at the same time/started during the 10-week study period	NI Possible cointerventions include melatonin, sleep hygiene and/or behavioural techniques. However, there is no information in the paper about whether or not participants used such cointerventions	NI – no data on possible co-interventions were provided NI
4.2. Were numbers of switches to other interventions low?	NI	See above	NI – as above NI
4.3. Was implementation failure minor?	NI	NI There is not enough information about implementation fidelity in the paper (e.g. no detail on compliance in taking the capsules)	NI – there is not enough information about implementation fidelity in the paper (e.g. there is no detail on compliance in taking the capsules) NI
4.4. If N or PN to 4,1, 4.2 or 4.3: Were adjustment techniques used that are likely to correct for these issues?	NI	See above	NI – as above NI
Bias because of missing data			
5.1 Are outcome data reasonably complete?	NI There were no data about missing data	NI There is little detail in the paper, other than that 40 participants received the intervention. It is not clear if the 40 participants were the original 40 recruited or if there were more recruited but only 40 proceeded to take the intervention. There is no detail on if there were incomplete outcome data	NI – no information regarding missing data is provided NI

continued

TABLE 49 Summary of quality assessment for controlled before-and-after studies using ACROBAT-NRSI (*continued*)

Other non-pharmacological intervention (Yehuda *et al.*[160])			
Bias because of confounding			
Domain	**Reviewer 1 quality appraisal**	**Reviewer 2 quality appraisal**	**Reviewers 3 and 4 quality appraisals**
5.2 Was intervention status reasonably complete for those in whom it was sought?	Y It is clear who was in which group	See above	NI – as above NI – insufficient information
5.3 Are data reasonably complete for other variables in the analysis?	NI There were no data about missing data or other variables	NI There is no detail about whether or not any participants were excluded from the analysis	NI – there were no data about missing data or other variables
5.4 If N or PN to 5.1, 5.2 or 5.3: Are the proportion of participants and reasons for missing data similar across interventions?		See above	See above
5.5 If N or PN to 5.1, 5.2 or 5.3: Were appropriate statistical methods used to account for missing data?		See above	See above
Bias in measurement of outcomes			
The outcomes for this study are as follows: co-operation, good mood, ability to concentrate, fatigue during the day, preparing homework, quality of sleep and haemoglobin			
6.1 Was the outcome measure objective?	N All measures except the haemoglobin level were based on self-completed questionnaire	N For all measures except the haemoglobin test	N All outcomes were based on self-completed questionnaire data, with the exception of the haemoglobin test, which was measured by a blood assay on the first day of the study and at the end of the 10-week study period

Other non-pharmacological intervention (Yehuda *et al.*[160])			
Bias because of confounding			
Domain	**Reviewer 1 quality appraisal**	**Reviewer 2 quality appraisal**	**Reviewers 3 and 4 quality appraisals**
6.2 Were outcome assessors unaware of the intervention received by study participants?	N Assume that the participants were blind to the treatment group (owing to identical capsules). If so, then as the participant assessed their own outcomes, then they were unaware of intervention received. It is not clear if the person who took and measured the haemoglobin level was blinded	N Data were collected by self-report of participants and analysed/assessed by researchers. Neither group were blinded/ unaware of the intervention received	N No information regarding blinding was provided. The questionnaire was self-reported data for five or six outcomes. It was unclear if the haemoglobin measurement was undertaken by a blinded individual N – for all measures except haemoglobin level, as these were based on a self-completed questionnaire
6.3 Were the methods of outcome assessment comparable across intervention groups?	Y All outcomes were measured through self-completed questionnaires at day 1 and 10 weeks plus a haemoglobin test	Y	Y – all outcomes were measured through self-complete questionnaires at day 1 and 10 weeks plus a haemoglobin test Y
6.4 Were any systematic errors in measurement of the outcome unrelated to intervention received?	NI	Not sure how to judge this	NI – no information regarding errors were provided. For instance, reliability of haemoglobin measurement? NI

continued

TABLE 49 Summary of quality assessment for controlled before-and-after studies using ACROBAT-NRSI (*continued*)

Other non-pharmacological intervention (Yehuda *et al.*[160])			
Bias because of confounding			
Domain	**Reviewer 1 quality appraisal**	**Reviewer 2 quality appraisal**	**Reviewers 3 and 4 quality appraisals**
Bias in selection of the reported result			
Is the reported effect estimate unlikely to be selected, on the basis of the results, from . . . multiple outcome *measurements* within the outcome domain?	?NI All domains are reported but there is no protocol	NI The paper does not refer to a published protocol, and with little detail provided on the analysis (i.e. intended analysis and actual analysis), it is difficult to make a judgement. It is impossible to know whether or not there were multiple outcome measurements and whether or not those reported were in fact a subset (or not) of these	NI – agree with reviewer 2 NI
Is the reported effect estimate unlikely to be selected, on the basis of the results, from . . . multiple *analyses* of the intervention–outcome relationship?	?NI Two-way ANOVA reported	NI As above. With little information and no protocol, it is impossible to judge whether or not the analysis reported are a selective subset of a wider analysis or not	NI – agree with reviewer 2 NI
Is the reported effect estimate unlikely to be selected, on the basis of the results, from . . . different subgroups?	?NI No subgroup analysis	NI As above. With little information and no protocol, it is impossible to judge whether or not there were any subgroups analysed. No subgroup analysis is reported	NI – agree with reviewer 2 NI

ANOVA, analysis of variance; N, no; N/A, not applicable; NI, no information; PN, probably no; PY, probably yes; Y, yes.

Summary of quality assessment for before-and-after studies[64]

TABLE 50 Summary of quality assessment for before-and-after studies

Study	Were the selection/ eligibility criteria adequately reported?	Is the sample likely to be representative?	If yes, was it a random sample?	Were patients recruited prospectively?	Were patients recruited consecutively?	Was the participation rate adequate (> 80% of those eligible)?	Was there ≥ 80% follow-up from baseline?
Questions 1–7 *Parent-directed tailored interventions*							
Austin *et al.*[123]	No Details of the sample were given, but they were not clearly stated in terms of whether or not these details were inclusion or exclusion criteria	No The sample size was very small – six parents of seven children (but it was described as a preliminary evaluation)	N/A	Unclear No detail is given to be able to assess this	Unclear No detail is given to assess this	Unclear There is no detail given in the paper about the numbers of eligible children it cannot be assessed whether or not there was > 80% participation rate	Yes 7/8 were followed up
Beresford *et al.*[21]	No The inclusion/exclusion criteria are not described	No Small sample (n =12)	N/A	Yes	Unclear No detail is given to assess this	Unclear There is no detail given in the paper about the numbers of eligible children so cannot assess whether or not there was a participation rate of > 80%	Unclear Follow-up loss is not reported so unable to assess this
Quine and Wade[146]	Unclear Inclusion criteria are reported but no exclusion criteria were reported	No Authors report that the sample was a 'highly selective group' owing to the bias towards males. They also compared the group to a previous sleep problem prevalence study that they undertook. From this, they noted that the sample had more sleep management problems and a longer duration of sleep problems than the prevalence study group, and that there was more marital unhappiness and maternal irritability than the prevalence study. In addition, the sample was selected from playgroups for preschool children, so although the ages of the sample are not reported they will be biased towards the younger ages	N/A	Yes	Unclear Participants were volunteers not referrals and so there was not really an 'order' to be consecutive from	Unclear There is no detail given in the paper about the numbers of eligible children so it cannot be assessed whether or not there was > 80% participation rate	Yes

continued

TABLE 50 Summary of quality assessment for before-and-after studies (*continued*)

Study	Were the selection/ eligibility criteria adequately reported?	Is the sample likely to be representative?	If yes, was it a random sample?	Were patients recruited prospectively?	Were patients recruited consecutively?	Was the participation rate adequate (> 80% of those eligible)?	Was there ≥ 80% follow-up from baseline?
Weiskop *et al.*[126]	Yes	No The age range of the sample is biased towards younger children. This was intentional as the authors felt that the intervention would be more suitable to younger ages. All but one were in specialist education services. This may make the sample quite specific in terms of representativeness. Furthermore, the sample was partly recruited through a disability newsletter, but there is no information given on who receives this newsletter and if it represents a specific subset of families	N/A	Yes	Unclear There is insufficient detail to make a judgement on this	Unclear There is no detail given in the paper about the numbers of eligible children so it cannot be assessed whether or not there was a > 80% participation rate	No 77% were followed up
Parent-directed non-tailored interventions							
Beresford *et al.*[21]	No The inclusion/exclusion criteria were not described	No Small sample ($n = 22$)	N/A	Yes	Unclear No detail is given to assess this	Unclear There is no detail given in the paper about the numbers of eligible children so it cannot be assessed whether or not there was a > 80% participation rate	No 70% (post intervention), 65% (12-week follow-up) and 78% (24-week follow-up) were followed up
Beresford *et al.*[21]	No The inclusion/exclusion criteria are not described	No Small sample	N/A	Yes	Unclear No detail is given to assess this	Unclear There is no detail given in the paper about the numbers of eligible children so it cannot be assessed whether or not there was a > 80% participation rate	No 69% were followed up at 12 weeks and 62% at 24 weeks
Bramble[149]	Unclear Inclusion criteria reported but exclusion criteria not reported	No Small sample of 15 participants	N/A	Yes	Yes	Unclear There is no detail given in the paper about the numbers of eligible children so it cannot be assessed whether or not there was a > 80% participation rate	Yes Reported that there were no dropouts but there were missing data for at least five children on one score and seven on another

Study	Were the selection/ eligibility criteria adequately reported?	Is the sample likely to be representative?	If yes, was it a random sample?	Were patients recruited prospectively?	Were patients recruited consecutively?	Was the participation rate adequate (> 80% of those eligible)?	Was there ≥ 80% follow-up from baseline?
Reed *et al.*[129]	Yes	Unclear There was some variability in some demographics (e.g. ethnicity), but some were very homogenous. The authors report that child care was offered to minimise selection bias	N/A	Unclear Some participants were recruited via a medical centre with a record review used but it is unclear in the paper whether or not this record review was used after prospective recruitment or to select and recruit participants	Unclear There is insufficient detail to make a judgement on this	Unclear There is no detail given in the paper about the numbers of eligible children so it cannot be assessed whether or not there was a > 80% participation rate	Yes 80% follow-up
Yu *et al.*[151]	Yes	No The sample was biased towards younger children and, other than autism, it excluded those with neurological conditions that could have affected sleep (e.g. epilepsy)	N/A	Yes	Unclear There is insufficient detail to make a judgement on this	Unclear There is no detail given in the paper about the numbers of eligible children so it cannot be assessed whether or not there was a > 80% participation rate	Yes 85% follow-up
Other parent-directed interventions							
Peppers *et al.*[155]	Yes	No Small sample ($n = 23$), demographics of only intervention group reported	N/A	Yes	Unclear	No	Yes

continued

TABLE 50 Summary of quality assessment for before-and-after studies (*continued*)

Study	Were the selection/ eligibility criteria adequately reported?	Is the sample likely to be representative?	If yes, was it a random sample?	Were patients recruited prospectively?	Were patients recruited consecutively?	Was the participation rate adequate (> 80% of those eligible)?	Was there ≥ 80% follow-up from baseline?
Other non-pharmacological interventions							
Guilleminault *et al.*[158]	No	Unclear Little detail on the sample is given	N/A	Unclear Participants were those referred to a clinic but it is not clear if they were recruited at the time of referral or retrospectively	Unclear No detail is given on this	Unclear There is no detail given in the paper about the numbers of eligible children so it cannot be assessed whether or not there was a > 80% participation rate	Yes *N* = 14. It is reported that 5/14 children responded to treatment. Non-responders to treatment described as five boys and four girls. The implication is that none was lost to follow-up
Oriel *et al.*[159]	Yes	Unclear Participants were recruited through letters sent home through local ASD support classrooms	N/A	Yes	Unclear	Unclear	Yes
Yu and Hong[161]	Yes	Unclear	N/A	Unclear	Unclear	Unclear	Yes

TABLE 51 Summary of quality assessment for before-and-after studies of non-pharmacological interventions

Study	Was loss to follow-up reported?	Were relevant prognostic factors reported?	Were other relevant confounding factors reported? (e.g. use of co-interventions)	Was an appropriate measure of variability reported?	Was there an appropriate statistical analysis?	Were there any other important limitations?
Questions 8–13 *Parent-directed tailored interventions*						
Austin *et al.*[123]	Yes	No	No	Yes	Yes	Yes
				It reports SDs	Repeated-measures *t*-tests	Small sample. Note: described as preliminary evaluation
Beresford *et al.*[21]	Unclear	Unclear	Unclear	Yes	Yes	No
	There is no detail in the report to assess this	There is little detail about the sample to assess this	There is little detail about the sample to assess this	It reports SDs	Appendix E notes that, owing to a very small sample size, tests of statistical significance were not applied	
Quine and Wade[146]	Yes	No	No	Yes	No	Yes
		Very little detail about the sample is reported, including possible prognostic factors	Very little detail is available on the sample	It reports SDs	Descriptive before-and-after data are reported, but a test of difference is reported for some but not for all variables. It is not always clear what type of test was used	The multiple baseline design data collection was stopped [i.e. the plan was to collect diary data for all from time of entering the study (all at same time point)] but as the parents were tired and because some had to wait a long time to start the intervention, for most families, data were only collected for the first and second weeks at baseline. Therefore, it was not possible to compare baselines and interventions in 'real' time. The follow-up period immediately after the intervention is not reported, so it is not clear what the 'after' measurement relates to. A further 3-month follow-up was used but most of the data reported pertain to the first (unknown) follow-up Note: the authors say they used an age-matched control group for comparison but this is not reported in the results and only before-and-after results are reported

continued

TABLE 51 Summary of quality assessment for before-and-after studies of non-pharmacological interventions (*continued*)

Study	Was loss to follow-up reported?	Were relevant prognostic factors reported?	Were other relevant confounding factors reported? (e.g. use of co-interventions)	Was an appropriate measure of variability reported?	Was there an appropriate statistical analysis?	Were there any other important limitations?
Weiskop *et al.*[126]	Yes	No It is not clearly reported whether or not there were any comorbidities that could have influenced the outcome (e.g. if any participants experienced seizures or duration of sleep disturbance)	Yes The paper reports that some children were taking medication for behaviour and/or sleep problems	No	No The authors report that emphasis is placed on clinical significance and use of goal attainment scaling and visual analysis of graphs. There is no analysis of difference using a particular test, but perhaps this is correct given the small sample size ($n = 13$)	Yes Small sample ($n = 13$). Two studies were conducted (one with a sample of children with ASD and one with a sample of children with fragile X syndrome) and then combined 'in the interest of brevity of results'[126]. It is not clear why two separate studies were conducted and so it is difficult to establish the validity of combining the two. One study had a 12-month follow-up, whereas the other did not. In addition, the treatment duration varied for participants, with a minimum of 7 weeks, but it was longer if treatment was interrupted (e.g. because of illness)
Parent-directed non-tailored interventions						
Beresford *et al.*[21]	No The paper deals with missing data reported but not the characteristics of those lost to follow-up	No	No	Yes It reports SDs; CIs were plotted but not reported in text	Yes Repeated measures ANOVA, paired *t*-tests if appropriate, effect sizes reported	No
Beresford *et al.*[21]	No Procedures for dealing with missing data discussed (appendix E) but no details on characteristics of those lost to follow-up	No	No	Yes It reports SD; CIs were plotted but not reported in text	Yes Repeated measures ANOVA, paired *t*-tests	No

Study	Was loss to follow-up reported?	Were relevant prognostic factors reported?	Were other relevant confounding factors reported? (e.g. use of co-interventions)	Was an appropriate measure of variability reported?	Was there an appropriate statistical analysis?	Were there any other important limitations?
Bramble[149]	Yes	Yes Various descriptions of the sample, which may have influenced outcomes, are reported	No It is not reported whether or not children were using other sleep aids/ medications	Yes It reports SD or SE of the mean	Yes Comparisons of pre and post treatment, using Friedman and Wilcoxon signed-rank tests	Yes Some of results were not presented very clearly (e.g. there were graphs but no tables) There were some incomplete data and exclusions from analysis
Reed *et al.*[129]	Yes	Yes Children were excluded if they had factors likely to contribute to disordered sleep	Yes All medications taken by participants were reported	Yes It reports SDs	Yes Paired data, Wilcoxon signed-rank test	Yes Small sample size (20–25). Some actigraphy data were lost and so data from only 12 participants were used for this outcome measure
Yu *et al.*[151]	Yes	Yes Children with medical conditions that could have affected sleep were excluded. For the sample it is not clear what the duration of the sleep disturbance was prior to the study and thus whether or not this would have affected outcomes	Yes Medications use reported	Yes It reports SDs	Yes Repeated measures ANOVA	No
Other parent-directed interventions						
Peppers *et al.*[155]	Yes	Unclear Medication and comorbid diagnosis for the intervention group were reported	Unclear Medications used by intervention group were reported	Yes It reports SDs, SE and CIs	Yes *t*-tests and descriptive statistics	Yes

continued

TABLE 51 Summary of quality assessment for before-and-after studies of non-pharmacological interventions (*continued*)

Study	Was loss to follow-up reported?	Were relevant prognostic factors reported?	Were other relevant confounding factors reported? (e.g. use of co-interventions)	Was an appropriate measure of variability reported?	Was there an appropriate statistical analysis?	Were there any other important limitations?
Other non-pharmacological interventions						
Guilleminault *et al.*[158]	No It was not reported explicitly but can be deduced from the text that the study had data on response to treatment for all 14 participants	Yes Various sample descriptions were reported	Yes The use of other drugs was reported and standardised	Yes It reports SDs	Yes Repeated measures ANOVA	Yes Chloral hydrate was being used concurrently but withdrawn over several weeks as a sleep pattern was established. There was no discussion of whether or not this would introduce bias Small sample of 14 children Not much detail was given on the analysis and measures: sleep logs were used (TST, distribution of sleep bouts, longest wake and sleep periods), and actigraphy, but only for some participants, although it is not clear how many. Later, the authors report that sleep logs were not being regularly kept in two cases, indicating that there were unreliable data
Oriel *et al.*[159]	Yes No loss to follow-up	Unclear	Unclear	No	Yes	Unclear Researchers were unable to stop children from engaging in aquatic exercise in the control phases. Researchers could encourage parents to avoid these activities but had no control over what they actually did

Study	Was loss to follow-up reported?	Were relevant prognostic factors reported?	Were other relevant confounding factors reported? (e.g. use of co-interventions)	Was an appropriate measure of variability reported?	Was there an appropriate statistical analysis?	Were there any other important limitations?
Yu and Hong[161]	No	N/A	Unclear	Unclear	Unclear	No
	100%	But there was none	Patients who used other drugs for expectant treatments were not eligible. Interventions involved two processes – it is not clear on the relationship/interactions between the two	It reports mean and SDs	Not clear what statistical tests were used	

ANOVA, analysis of variance; N/A, not applicable.
Yes, low risk of bias; unclear, unclear risk of bias; no, high risk of bias.

Appendix 22 Parent sleep-related outcomes for studies evaluating non-comprehensive parent-directed interventions

TABLE 52 Parent sleep-related outcomes for studies evaluating non-comprehensive parent-directed interventions

Study	Parent sleep-related outcome assessed	Method of assessment	Definitions[a]
Global measures and composite scores			
Wiggs and Stores[152]	TST	Actigraphy	Time from sleep onset to the time the child woke up
Parent-carer-related quality of life			
Wiggs and Stores[152]	Parental stress	Malaise Inventory, reported separately for mothers and fathers	The Malaise Inventory is a 24-item binary choice questionnaire that has been used in a number of studies of disabled children to measure carers' stress. Scores of 5 or 6 are considered to indicate stress outside the normal range. Scores of ≥ 7 are said to have critical implications for physical and mental health
Sleep maintenance			
Wiggs and Stores[152]	Parent sleep (sleep period, activity score, movement index and fragmentation index)	Activity monitors	Sleep period: time from sleep onset to the time the child woke up. Referred to as sleep period Activity score: the sum of the epoch scores divided by total number of epochs in the sleep period Movement index: the number of thirties epochs with a value > 0 (i.e. with movement), divided by the total number of thirties epochs in the sleep period × 100 Fragmentation index: the number of discrete thirties epochs with a value of 0 (i.e. with no movement) divided by the total number of immobile phases of any duration × 100
Sleep scheduling			
Wiggs and Stores[152]	Daytime sleepiness (mothers and fathers)	Epworth sleepiness scale	An 8-item self-report scale concerning the likelihood of falling asleep in everyday situations. Sleeping propensity was measured on a 4-point scale and scored as follows: 0 = would never sleep, 1 = slight chance of sleeping, 2 = quite likely that they would sleep, 3 = very likely that they would sleep. The possible score range was from 0 to 24. Scores of ≥ 11 may indicate hypersomnolescence. The authors provide a reference

continued

TABLE 52 Parent sleep-related outcomes for studies evaluating non-comprehensive parent-directed interventions (*continued*)

Study	Parent sleep-related outcome assessed	Method of assessment	Definitions[a]
Quality of sleep			
Wiggs and Stores[152]	Parental satisfaction with own sleep	A six-point Likert scale, reported separately for mothers and fathers	A six-point Likert scale; the responses were totally satisfied, satisfied but could be better, more often satisfied than not satisfied, more often unsatisfied than satisfied, unsatisfied but could be worse and totally unsatisfied
Other outcomes			
Wiggs and Stores[152]	Parental satisfaction with child's sleep and parental satisfaction with their ability to cope with their child's sleep pattern and daytime behaviours	A six-point Likert scale, reported separately for mothers and fathers	A six-point Likert scale; the responses were totally satisfied, satisfied but could be better, more often satisfied than not satisfied, more often unsatisfied than satisfied, unsatisfied but could be worse and totally unsatisfied

a As defined by study authors.

Appendix 23 Measures of perceived confidence and/or efficacy and/or understanding of sleep/sleep management for studies evaluating non-comprehensive parent-directed interventions

TABLE 53 Measures of perceived confidence and/or efficacy and/or understanding of sleep/sleep management for studies evaluating non-comprehensive parent-directed interventions

Study	Method of assessment	Definitions[a]
Behavioural		
Peppers *et al.*[155]	Parent Satisfaction Likert Survey	No further definition or reference provided
Wiggs and Stores[152]	The parents' perceived control to manage own and partners' sleep difficulties (VAS), reported separately for mother and father	VAS (100 mm) scored from 0 to 100. Parents rated their ability on a scale of not at all able to control it to totally able to control it. Parents also rated their partners
	The parents' locus of control: internally/externally control scale reported separately for mother and father	A 29-item forced choice questionnaire that includes six filler items to make the test purpose ambiguous. It was used to measure parents' orientation to internal or external beliefs. External control beliefs: events are caused by some attribute of the environment including powerful others, fate or luck. Internal control beliefs: events are contingent on own actions or own relatively permanent characteristics. Individuals scoring highly on an internal belief scale tend to be more resilient to negative events and are less likely to develop subsequent psychological or physical characteristics. The authors provide a reference

VAS, visual analogue scale.
a As defined by study authors.

EME
HS&DR
HTA
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