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Evidence on Effectiveness of Behavioural Interventions to Help Parents Manage Sleep Problems in Young Disabled Children: A Rapid Review

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1. Introduction

Sleep problems are common among all children but they appear to be more common among disabled children. For example, Quine[1] found that settling problems were reported for 41 per cent of children aged 4-12 in special schools compared with 27 per cent of children in mainstream schools; figures for night waking were 45 per cent compared with 13 per cent. Figures for children with severe learning disability are particularly high: for example, Bartlett *et al.*[2] reported problems in over 80 per cent of children aged up to 11 years and 77 per cent of 12 to 16 years and Richdale and Prior[3] reported prevalence of 34-80 per cent in children with autism. Such problems appear to be very persistent. For instance, Wiggs and Stores[4] showed average duration of current sleep problem was 7.13 years, and problems are not likely to disappear without intervention.[5]

A number of reasons have been suggested for the high prevalence of sleep problems in disabled children. Physical and medical conditions associated with disability may impact on sleep[1]. This can be particularly the case for technology dependent children. Recent research on the experiences of families of children dependent on medical technology shows that sleep disturbance for the child and parents is common due to the need to attend to technology, such as feeding pumps or dialysis machines, during the night, and to machine alarms going off frequently.[6] Problems in cognition and learning can hinder the establishment of appropriate routines for settling and staying asleep and parents may also have low expectations of the child's ability to learn such routines.[7]

Sleep problems have a number of implications for the child and family. For parents, they are associated with high levels of stress and irritability.[8] For the children they are associated with poor concentration and daytime learning, and increased probability of daytime behaviour problems.[5] These findings emphasise the need to take sleep problems seriously. However, only a minority of families who have a child with a severe sleep problem appear to receive any help.[4]

In considering whether intervention is needed, it is important to note that it is normal for young children to wake a number of times during the night.[9,10] What distinguishes normal sleep from a sleep problem is what children do when they awaken. In normal sleep, children wake briefly and resume sleep themselves (self-settling). Children with sleep problems signal when they wake and elicit a response from parents, this can act as a reward and result in the child needing parental attention to resume

sleep. As France *et al.*[9] note 'intervention does not involve changing the child's sleep per se ... but involves teaching the child to replace the behaviour of signaling upon awakening with the behavioral quietude necessary for the resumption of sleep' (p.583). Young children also often spend some time settling themselves to sleep when put to bed. However this becomes a problem when a child makes repeated calls on parents after being put to bed. Again the aim of intervention is to teach the child to fall asleep alone.

Sleep problems encountered in studies of disabled children are broadly of two types: a) 'behavioural' problems relating to the initiation and maintenance of sleep, as described above, and linked to parental management; and b) 'physical' problems, such as upper airway obstruction and other physiological factors. However, these often co-exist, and it is important that a full assessment of the problems and their causes is carried out to inform the choice of intervention. Stores and Wiggs[11] suggest that questions regarding the child's sleep-wake patterns should be a routine part of any general assessment. They recommend the following screening questions:

1. Does the child have any difficulty getting to sleep or staying asleep?
2. Is the child excessively sleepy/over-active during the day?
3. Does the child have any disturbed episodes at night?

Positive answers to these questions should lead to a detailed investigation, including sleep history and physical examination, and choice of interventions should be individually tailored to the child's problems.[11]

This rapid review focuses on interventions for behavioural sleep problems in young disabled children (up to age eight years), specifically interventions that can be carried out by parents in the home.

2. Methods

A rapid review was undertaken on the effectiveness of behavioural interventions for sleep problems in disabled children.

2.1 Searches

The search was structured to combine the following concepts:

> Sleep problems AND (children terms in close word proximity to disabled terms) AND behavioural interventions

Case studies, letters, notes, comments and editorials were excluded from the searches. Searches were restricted to English language studies published since 1985. The full search strategies are reported in Appendix A.

A range of databases and websites were searched (see Table 1). Records were downloaded and added to Endnote bibliographic software. The records were deduplicated.

Table 1: Databases searched for research evidence on behavioural interventions for sleep problems in disabled children

Database	Interface	Date searched
Cochrane Database of Systematic Reviews (CDSR)	Cochrane Library 2008 Issue 3	22/8/2008
DARE	Cochrane Library 2008 Issue 3	22/8/2008
MEDLINE	Ovid MEDLINE(R) In-Process & Other Non-Indexed Citations and Ovid MEDLINE(R) <1950 to Present>	22/8/2008 22/9/2008 (Revised search)
EMBASE	OvidSP, 1980 to 2008 Week 33	22/8/2008
PsycINFO	OvidSP,1967 to July Week 5 2008	22/8/2008 22/9/2008 (Revised search)

Database	Interface	Date searched
CINAHL	OvidSP, 1982 to August Week 3 2008	22/8/2008
CENTRAL	Cochrane Library 2008 Issue 3	22/8/2008
SPECTR and C2-RIPE (Campbell Collaboration)	http://geb9101.gse.upenn.edu	22/8/2008
HMIC	Ovid to July 2008	22/8/2008
NRR archive	https://portal.nihr.ac.uk/Pages/NRRArchiveSearch.aspx	22/8/2008
CERUK	http://www.ceruk.ac.uk/	22/8/2008
ERIC	Dialog/Datastar	22/8/2008
Childdata	http://www.childdata.org.uk/library_search.asp	26/8/2008
Australian Education index (AUEI)	Dialog/Datastar	29/8/2008
British Education Index (BRIE)	Dialog/Datastar	29/8/2008

2.2 Inclusion and exclusion criteria

For the review of sleep interventions two researchers independently screened titles and abstracts. Full papers were ordered for any records identified by either researcher as potentially relevant. These were also screened by two researchers based on the criteria below (Table 2). Any disagreements were resolved by discussion and a consensus decision was made.

Table 2: Inclusion and exclusion criteria

Exclusion criteria

- Not English language
- Published before 1985
- Research not concerned with intervention to manage/address/resolve a sleep problem
- Pharmacological intervention only
- Interventions other than those adopting a behavioural approach
- Interventions which only and specifically address the following sleep problems:
  - night terrors
  - sleep walking
  - sleep apnoea
- Research does not include any evaluative element
- Research where the sample includes disabled and non-disabled children, and no separate analysis
- Case studies, letters, notes, editorials
- No quantitative outcome measures used
- Age of sample 9 years or older (inclusive)
- Sample only includes children with the following as their 'primary need':
  - attention deficit hyperactivity disorder (ADHD)
  - mental health problems
  - emotional/social/behavioural difficulties

Inclusion critieria

- Intervention includes at least a behavioural intervention element to manage/address/resolve a sleep problem

and

- Intervention for disabled children aged 8yrs and under

and

- Evaluation of that intervention which includes, at least, a quantitative element

During screening, it became apparent that an age cut-off of eight years old was not commonly used by studies. We made the decision to included studies including children older than eight provided they included a substantial proportion of children who were our population of interest (i.e. young children under eight).

2.3 Data extraction

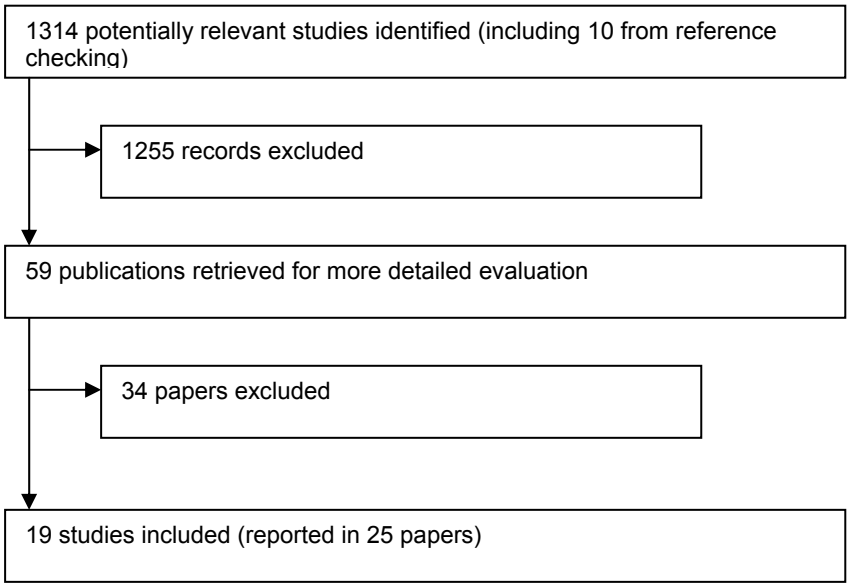
Data were extracted into a standardised form (see Appendix D) by one researcher. A sample of four sets of data extraction was checked by a second researcher. Study design was classified according to the Maryland Scale of Scientific Methods.[12] Studies with a control/or comparison group were also quality appraised using criteria from the Effective Public Health Practice Project Quality Assessment Tool for Quantitative Studies.[13]

3. Results

3.1 Study selection

1,314 records were screened for relevance, 1,304 from the electronic searches and 10 publications identified through reference checking and other sources (see Figure 1). 1,255 records were excluded and 59 publications were retrieved for more detailed evaluation. Twenty-five papers met the inclusion criteria for the review reporting on 19 individual studies. Thirty-four papers were excluded. See Appendix B for list and reasons for exclusion.

Figure 1: Study selection



3.2 Overview of included studies

The included studies have been grouped by type of intervention (Table 3). Six studies (n=239) evaluated a non-specific behavioural intervention i.e. they did not focus on a single behavioural technique;[14-19] seven evaluated extinction or graduated extinction (n=48);[20-26] two evaluated sleep restriction (n=6);[27,28] and three evaluated faded bedtime with response cost (n=21).[29-32] Full details of one study (n=5) (available in an MSc thesis) had not arrived at the time of writing therefore this study is not discussed any further.[33]

Based on the Maryland Scale of Scientific Methods, only study designs at Level 3 to 5, which encompass various study designs with a control or comparison group, are sufficient to inform whether an intervention works, does not work or is promising. Only four of the 19 studies met the criteria for Level 3 or above on the Maryland scale: three were of a non-specific behavioural intervention[14-16] and one was of faded bedtime with response cost.[29] The remaining studies were all before and after design and did not have a control group. When evaluating whether or not an intervention works the absence of a control group is a key limitation as it is not possible to rule out with any certainty the possibility that factors other than the intervention may have led to change. However, in the absence of any better quality available evidence, details of these studies are provided below as they provide potentially useful information on acceptability of different interventions and the feasibility of using them with different groups of disabled children.

Some caution also needs to be taken when considering how the findings of any of the studies included in this review might be generalised to other disabled children with sleep problems. A key question is whether the parents who participated in a particular study are representative of parents of disabled children with sleep problems. Such parents may differ in many ways. For example, there is the possibility that parents who participate in such studies are more highly motivated and/or feel more confident about dealing effectively with their child's sleep problems and/or are at a stage where they can feel they can take such an intervention on. In this instance it is possible that when delivering this intervention outside the context of a research study that the results may not be as good.

Table 3: Overview of included studies

Author	Year	Study design Maryland Level	Number of participants	Intervention	Comparator	Country and setting
Non-specific behavioural intervention						
Montgomery[14]	2004	RCT Level 5	N=66	(a) Behavioural intervention (BI) delivered to parents face-to-face (b) BI delivered through a booklet	Waiting-list control	UK Home
Stores[15]	2004	RCT Level 4	N=46	Single session of instruction on behavioural techniques plus booklet	Waiting-list control	UK Home
Wiggs[16] Related publications Wiggs[33] Wiggs[34]	1998	RCT Level 4	N=31	Tailored BI	Waiting-list control	UK Home
Bartlet[17]	1998	BA Level 2	N=61	Tailored BI (mainly graded change)	No	UK Home
Hewitt[18]	1985	BA Level 2	N=10	Tailored BI (positive bedtime routine and conditioning)	No	UK Home
Quine[19] Related publications Quine[35] Quine[36] Quine[37]	1991	BA Level 2	N=25	Tailored BI (positive bedtime routine and conditioning)	No	UK Home
Extinction						
Bramble[20] Related publications Bramble[38]	1996	BA Level 2	N=15	Extinction	No	UK Home
Didden[21]	2004	BA Level 2	N=3	Extinction (n=2); differential reinforcement of incompatible behaviours plus response cost (n=1)	No	Netherlands Home
Didden[22]	2002	BA	N=4	Extinction	No	Netherlands

		Level 2				Home
Didden[23]	1998	BA Level 2	N=6	Extinction	No	Netherlands Home
Durand[24]	1996	BA Level 2	N=4	Graduated extinction	No	USA Home
Thackery[25]	2002	BA Level 2	N=3	Extinction with positive bedtime routine	No	Australia Home
Weiskop[26]	2005	BA Level 2	N=13	Extinction with positive bedtime routine	No	Australia Home
Sleep restriction						
Christodulu[27]	2004	BA Level 2	N=4	Positive bedtime routine and sleep restriction	No	USA Home
Durand[28]	2004	BA Level 2	N=2	Positive bedtime routine and sleep restriction	No	USA Home
Faded bedtime with response cost						
Piazza[29]	1997	RCT Level 4	N=14	Faded bedtime with response cost	Bedtime scheduling	USA Inpatient
Piazza[30]	1991	BA Level 2	N=3	Faded bedtime with response cost	No	USA Inpatient
Piazza[31]	1991	BA Level 2	N=4	Faded bedtime with response cost	No	USA Inpatient
Unclear						
Colville[32]	1996	BA Level 2	N=5	BI (details not provided)	No	UK Home

BI: behavioural intervention, BA: before and after study design, RCT: randomised controlled trial.

3.3 Non-specific behavioural intervention

All six studies of a non-specific behavioural intervention were conducted in the UK and the intervention was delivered by parents to their children in their own home. The age range of children varied between studies (Table 4). With the exception of one study that included children with a chronic illness,[17] the majority of participants had learning disabilities which were mainly severe. One study, with the objective of assessing the effectiveness of a simple behavioural approach for prevention as well as minimisation of sleep problems, included children with and without sleep problems.[15] The remaining five studies used different methods to assess the severity of the children's sleeping problems at baseline making it difficult to be certain about the similarity of the populations across the study. However, overall the children appear to have had severe sleep problems which were predominantly long-standing. The most commonly reported problems were difficulties in settling at bedtime and related disruptive behaviour, several episodes of night waking leading to disrupted sleep for parents and other members of the household and co-sleeping.

Table 4: Details of participants (non-specific behavioural interventions)

Study (N)	Disability	Age	Baseline severity of sleep problem
Randomised controlled trials			
Montgomery[14] Face-to-face n=20 Booklet n=22 Control n=24	Severe LD	Range 2-8 years	Severe sleep problem (CSDS score ≥4) was an entry requirement CSDS mean 6.55 (SD 1.31)
Stores[15] N=46	Down Syndrome (severity of LD not stated)	Mean 2yr 8mth Range 7mth – 4yr 9mth	65% had at least one behavioural sleep problem; 35% did not have a sleep problem
Wiggs[16] Intervention n=15† Control n=15	Severe LD (with ≥1 daytime challenging behaviours)	I mean 8.2 (SD 2.7) C Mean 10.8 (3.8)	Severe sleep problem was an entry requirement
Before and after studies			
Bartlet[17] n=61	N=22 chronic illness; n=39 disability (most commonly severe LD)	Mean 4yr 11mth Range 11mth-17yr	SDI score mean 6.36

Study (N)	Disability	Age	Baseline severity of sleep problem
Hewitt[18] n=10	Severe LD	Mean 6yr 11mth Range 3yr 11mth-16yr 6mth	Average time to settle ranged from 34min to 2.5hr; 6 to 28 night waking episodes in one week
Quine[19] n=25	Severe LD	Range 1yr 9mth to 21 yrs	Mean time to settle 111 min (range 45-180); mean 3.1 times waking per night (range 2.2-4.0)

LD: learning disability, CSDS: Composite Sleep Disturbance Score (ranges from 0 to 8, higher score more severe problem), SDI: Sleep Disturbance Index (ranges from 0 to 8, higher score more severe problem), I: intervention group, C: control group, † There were n=16 allocated to the intervention but one dropped out before receiving the intervention.

Although all six studies were similar in that they provided parents with information on more than one behavioural technique, they did vary in how the intervention was implemented (Table 5). Two RCTs[14,15] provided single general information sessions for parents on behavioural techniques and one RCT[16] and the before and after studies[17-19] provided individual treatment plans for each child based on a functional assessment.

3.3.1 General information sessions

Montgomery *et al.*[14] evaluated the effectiveness of (i) a single information session on behavioural interventions delivered to parents face-to-face in their own home and (ii) information on behavioural interventions delivered through a booklet. There were 20 participants in the face-to-face group, 22 in the booklet group and 24 participants in a waiting list control group (Table 4). The aim was to train parents in both the face-to-face and booklet groups in the same behavioural techniques (see Box 1). At baseline participants in all the groups completed a sleep questionnaire and kept a sleep diary for two weeks. The intervention groups then received a 90 minute visit from a researcher to explain the behavioural techniques (face-to-face) or received a 14 page illustrated booklet providing the same information (Table 5). The intention was that parents would then implement the techniques with their children over a six week period.

Box 1: Information on behavioural techniques provided to parents in Montgomery et al. study14

a) Explanation of the benefits of normal sleep
b) Introduction to behavioural techniques in general (e.g. how behaviours can be triggered by preceding events, ignoring and consistency)
c) Recording behaviour in a sleep diary to devise and monitor treatment plans
d) Good sleep habits (e.g. clear routines, putting children to sleep while drowsy)
e) Techniques for changing settling and waking problems (ignoring child, checking and briefly at increasingly longer intervals and with minimal contact)
f) Removing child from parental bed using the settling techniques above
g) Rewards for desirable behaviour

The primary outcome measure was the Composite Sleep Disturbance Score (CSDS) which scores duration and frequency of settling and waking problems based on sleep diaries completed by parents. The possible score range is from 0 to 8 with a higher score indicating greater sleep problems. At baseline the mean score was six or greater for both intervention groups and the control group (see Appendix D for full data).

There was a statistically significant improvement for both of the intervention groups compared to the control group at end of treatment. Post-treatment the mean CSDS was 2.4 (SD 1.93), 2.55 (SD 2.76) and 5.75 (SD 1.54) for the face-to-face, booklet and control group respectively. This improvement was maintained for the two intervention groups at six month follow-up.

Prior to the intervention, parents were asked what minimum improvement would make the intervention worthwhile: 83 per cent said that having the problem reduced by half would make it worthwhile. Based on this a positive treatment response (responder) was defined as a reduction of at least 50 per cent on the CSDS. Based on this classification there were 15 responders and five non-responders in the face-to-face group; 15 responders and seven non-responders in the booklet group; and all non-responders in the control group. Parents who had used the booklet were asked to rate its usefulness, ease of understanding and relevance. On a rating scale with a maximum score of 12 the mean score was 10.17 (SD 1.87).

This was a good quality RCT with a low risk of bias (see Appendix C for full quality assessment) therefore the findings are likely to be reliable. There are two key points that need to be kept in mind when interpreting the findings. Firstly, as emphasised by the authors the study was not designed to directly compare the effectiveness of delivery of information face-to-face with delivery face-to-face. It is not powered (i.e. does not have enough participants) to detect whether one mode of delivery is more effective than the other: it assesses whether each of the interventions is better than no intervention. Secondly, the booklet group also (in common with the face-to-face group) had a total 90 minutes one-to-one contact with the researchers throughout the duration of the study for the purpose of assessing progress. This contact may have had a supportive and motivational value for parents and it is possible that this contact may have contributed to the effectiveness of the booklet intervention. Further work is required to unravel the contribution of the booklet and the contact with researchers/clinicians. In terms of generalising the findings, it is possible that providing a booklet, outside the research context with no regular contact with the clinical team, may not be as effective as in this study.

Stores and Stores[15] compared a single session of instruction on behavioural techniques plus provision of a booklet to a waiting list control group. Forty-six children were randomised to either the intervention group or control group. (The number of participants in each group was not explicitly stated.) The instruction session lasted approximately 90 minutes including 30 minutes for discussion and was delivered to small groups of about five mothers. There were separate sessions for mothers of very young children (six months to 2.5 years old) and young children (2.5 to five years old). The session included provision of information and advice about children's sleep and explanation of behavioural techniques for encouraging good sleep habits such as establishing a positive bedtime routine, rewarding good behaviour, ignoring unwanted behaviour and gradual change. Case studies were used to illustrate the techniques. The intention was that parents would then implement the techniques with their children over a four week period.

Sixty-five per cent of the children had at least one behavioural sleep problem and 35 per cent did not have any sleep problems. On the Composite Sleep Problem Score (CSPS) with a possible score range of 0 to 14 (a higher score indicated worse sleep

problems) the mean baseline score for the intervention and control group was 3.83 (SD 3.41) and 3.38 (SD 3.58) respectively. Based on a three (baseline, one month and six month follow-up) by two (intervention and control group) analysis of variance there was no statistically significant effect for time or group or interaction between group and time. There was a statistically significant difference between the intervention and control group at six months based on a post-hoc test; however this should be treated with caution as, in the absence of any statistically significant differences based on the ANOVA, this may be a spurious finding.

The study also assessed the impact of the intervention on mothers' knowledge as assessed by two questionnaires. At one month follow-up, mothers in the intervention group scored more highly than the control group on the Knowledge of Behavioural Principles as Applied to Children Questionnaire and the Knowledge of the Sleep of Young Children Questionnaire and the differences were statistically significant (see Appendix D for complete data). Ninety-four per cent rated the information session and booklet as very easy to understand. Twenty-two per cent rated the presentation as 'very useful' and 61 per cent as 'quite useful'; 17 per cent rated the booklet as 'very useful' and 50 per cent as quite useful; the remaining participants gave a rating of 'not very useful'.

Although this study was an RCT, the use of a mixed group of children with and without sleeping problems limits how informative it is about the effectiveness of behavioural interventions for children with sleep problems. The aim of the study was to investigate the usefulness of the intervention for the prevention of sleep problems as well as treatment. Because the data from children with and without sleep problems was analysed as one group the mean severity of sleep problems at baseline was fairly low. As a result there was limited room for improvement on the scale that was used (i.e. a ceiling effect). It is therefore not surprising that there was not a statistically significant difference between the two groups in the main analysis.

The Maryland criteria require at least two Level 3 evaluations showing effectiveness to classify an intervention as effective and one Level 3 evaluation to classify an intervention as promising. Based on these criteria, the provision of information on behavioural techniques to parents in a single session (face-to-face) or through a

booklet is a promising intervention for dealing with severe behavioural sleep problems in children with learning disabilities.

3.3.2 Individual treatment plans

One RCT[16] and three before and after studies[17-19] provided individual behavioural treatment plans for each child based on a functional assessment. Wiggs and Stores[16] compared a tailored behavioural intervention received by 15 children (see Box 2) to a waiting list control group of 15 children. The children had severe learning disabilities and one or more daytime challenging behaviours (see Table 4 and 5). Only children with a severe sleep problem were included in the study.

Following an introductory visit to meet parents at home and explain baseline questionnaires there was a second visit to undertake a functional analysis of the sleep problem and to agree the behavioural programme. This visit lasted between 1.5 and 2.5 hours. The functional assessment was based on sleep diaries completed by parents and a semi-structured interview to take a detailed sleep history. During this visit there was also discussion of possible factors maintaining their child's sleep problem as well as discussion of the positive and negative aspects of different behavioural techniques that might be useful. The techniques discussed included extinction, graded extinction, stimulus control procedures and positive reinforcement. The aim was to enable parents to make an informed choice about whether they would be able to implement a particular technique with their child. A behavioural programme was agreed with parents and following the visit they were sent a written outline of the agreed programme. The intention was that parents would then implement the agreed programme with their children over a four week period. Progress was monitored by regular telephone calls. Both the intervention and control group received the preliminary visit and four visits to deliver and collect questionnaires.

Box 2: Summary of a tailored behavioural intervention (Wiggs and Stores)[16]

a)	Functional analysis of child's sleep problem
b)	Establish what the parents' aims of treatment were
c)	Discussion of factors and mechanisms that maintain the child's problems in settling and or night-waking
d)	Discussion of different behavioural techniques, their advantages and disadvantages and how they might be applied to the specific family situation
e)	Identification and anticipation of any problems that might arise with the intervention
f)	Identification of target/s for the first stage
g)	Written outline of the agreed behavioural programme sent to parents following the visit

Outcome was assessed at the end of the four week intervention (one month follow-up) and three months following the commencement of treatment (three month follow-up). Nine groups of child and parent-related outcomes were reported.

In terms of child sleep problems the intervention group showed a statistically significant improvement from baseline to one month follow-up and baseline to three month follow-up on the Composite Sleep Index (CSI), whereas there was no change in the control group (see Appendix D for details of analysis). The CSI had a possible range of 0 to 12 with a higher score indicating greater severity. The mean score reduced from 6.73 (SD 2.31) at baseline to 3.79 (SD 1.89) and 2.96 (SD 2.24) at one and three month follow-up respectively. The mean CSI score for the control group for the same time periods was 7.23 (SD 2.26), 6.62 (SD 1.89) and 6.29 (SD 2.60). There were no between group differences in change in child sleep over time as measured by a wristwatch activity monitor. Also there was no change in daytime behaviour measured by the Aberrant Behaviour Checklist completed by mothers and teachers or in the severity and frequency of target challenging behaviours again assessed by mothers and teachers.

Several parental outcomes were assessed. There was a statistically significant increased sleep period (as measured by a wristwatch activity monitor) for mothers in the intervention group, from baseline to one month follow-up, compared to control

(see Appendix D). Mother and father satisfaction with their own sleep and their child's sleep also improved from baseline to one month and three month follow-up for the intervention group compared to control. There was also increased satisfaction amongst intervention mothers in how they coped with their child's sleep pattern, though no difference in how they rated their ability to control their child's sleep-related problems. Mothers in the treatment group reported reduced stress (The Malaise Inventory) from baseline to three month follow-up compared to control. There were no between group differences for fathers' stress. Based on the Internal/External Locus of Control Scale there was an increase in treatment group fathers' externality and a reduction for the control group. There was no statistically significant between group differences for mothers.

Although this study was an RCT it does have some limitations which may introduce the risk of over-estimating the effectiveness of the intervention (see Appendix C for full quality assessment). Randomisation was by school rather than individual child to avoid contamination. While this can be an appropriate way to avoid contamination, details of the methods were not reported, for example the number of schools randomised was not reported therefore it is unclear how many clusters there were. Additionally, the method of statistical analysis does not seem to have taken into account the clustering effect within schools in terms of characteristics such as type of disability, severity of disability or social background.

The three before and after studies used a similar tailored intervention to that of Wiggs and Stores[16] above (see Table 6) with 10,[18] 25,[19] and 61[17] participants. In particular, the treatment approaches described by Quine and Wade and Hewitt were very similar (see Box 3). Bartlet and Beaumont do not provide a detailed report of their intervention, from the information provided they appear to have taken a similar approach.[13] They report that the most commonly used strategies by parents were cueing, graded change, extinction and positive reinforcement.

Box 3: Intervention used by before and after studies (Quine and Wade[19] and Hewitt[18])

a) Positive bedtime routine including set bedtime and avoidance of overstimulation in the hour before bed; a regular routine providing clear stimuli for the child that bedtime is approaching
b) Teaching a relaxation response after getting into bed through use of a bedtime story or soft music
c) Gradual distancing of parent from bedroom once relaxation response was established
d) Identification of factors that were maintaining disruptive behaviours and advice for more constructive parental responses
e) During wakeful episodes the stimulus being used to condition the child to fall asleep was repeated. Parents were advised to interact with the child as little as possible and avoid prolonged routines and overstimulation during waking episodes
f) Parents were made aware of the importance of consistency and the possibility that progress may be slow

There were one-to-one meetings with parents at home or in a clinic to introduce the study and to develop an individual treatment plan for each child. Although a range of behavioural techniques was used, positive bedtime routine with graded change was predominant. Support for parents was fairly intensive. There were weekly visits from a nurse or health-visitor initially in two studies (Table 5).[18,19] In the third study contact with parents was usually by telephone: on average five phone calls per family ranging in duration from five to 60 minutes.[17]

A key difference between the before and after studies and the RCT on individual behavioural treatment plans was that three before and after studies did not have a pre-specified duration of implementation. The intervention was implemented until parents were satisfied with the progress made (Table 5) and then the outcomes of interest were assessed. While this makes clinical sense, in terms of evaluating the effectiveness of an intervention it does make it more likely that a positive impact of an intervention will be found, particularly in the context of a before and after study. The study by Quine and Wade compared their cohort of participants to an age-matched random sample of children with sleep problems from another health district who had not sought or been offered treatment. However, the outcomes of the two groups

were not directly compared: before and after comparisons were made within each group, not between groups. This study was therefore classified as a before and after study though a summary of the outcomes for the control group was extracted (see Appendix D).

All three studies showed improvement on child sleep outcomes and the two studies assessing parental outcomes also showed positive changes post-intervention (see Appendix D for full details).[17-19]

The authors of the studies make a number of points of interest in relation to implementing behavioural sleep interventions in families with a young disabled child. Hewitt[18] highlights that many programme modifications were necessary to ensure that the individual interventions suited individual parenting styles and family resources.

Bartlet and Beaumont[17] described their experience during a one year project based at Southampton General hospital staffed by a part-time experienced health visitor and a child psychiatrist four hours per week. The authors comment that treatment was often found by the parents as being more onerous than the literature had previously suggested. Forty-five children improved following the intervention and seven parents found the programme difficult to manage or ineffective.

A preliminary intervention was required for approximately one third of parents prior to being trained in the behavioural techniques to be used with their child.[13] Particular issues for parents included physical exhaustion, disagreement between partners about the way forward, low self-esteem, and a concern that the child would suffer as a result of the intervention. Tearfulness and feelings of hopelessness were common and three mothers were identified as clinically depressed and were referred to their GP for help. The aim of the preliminary intervention with parents was to allow time to develop trusting relationships with the project workers and to give them time to think and contemplate changing their routines. Specific details of the preliminary intervention were not provided other than that a holistic, dynamic approach was used with strategies such as understanding, support, empowerment and opportunities to talk through past traumatic experiences.

This experience is of particular interest from this study as it is based on one year's experience at a clinic therefore the participants may be more representative of parents of disabled children than parents recruited into a research project.

Based on the Maryland Criteria, a behavioural intervention delivered through an individual treatment plan is a promising intervention for dealing with severe sleep problems in disabled children.

Table 5: Details of interventions (non-specific behavioural intervention studies)

Study	Details of intervention	Duration of implementation	Support for parents
Randomised controlled trials			
Montgomery[14]	(a) Face-to-face – 90 minutes single session to explain range of behavioural techniques (in individual homes) (b) Booklet – were provided with 14 page booklet explaining same behavioural techniques Range of behavioural techniques.	Six weeks	No support specified beyond the initial session to (a) explain the technique or (b) give booklet.
Stores[15]	Small group 90 minute single session to explain range of behavioural techniques. Separate sessions for mothers of under 2.5 year olds and 2.5 to 5yr olds. Also provided with booklet. Range of behavioural techniques.	One month	No support beyond single session.
Wiggs[16]	One-to-one meeting with parents at home (1.5 to 2.5hr duration) to undertake functional analysis and agree detailed behavioural programme. Written details of agreed programme sent to parents. Range of behavioural techniques.	One month	Progress was monitored by regular telephone calls.
Before and after studies			
Bartlet[17]	One-to-one meeting with parents at home or clinic (one or two appointments depending on needs). About one third of parents received a preliminary intervention prior to this before they were ready to become involved in the programme. Range of behavioural techniques. Graded change was used in a high proportion of cases.	Until parents were satisfied with the progress made. Generally three months.	Contact usually by telephone. Mean number of calls 4.95; duration ranged from 5 to 60min.

Study	Details of intervention	Duration of implementation	Support for parents
Hewitt[18]	One-to-one meeting with parents at home to agree behavioural programme (two appointments). Details written up for parents. Mainly positive bedtime routine and graded change. Tailored to individual needs.	Until parents were satisfied with the progress made. Mean 6.7 weeks; range 2-15.	Weekly visits from nurse and visits from psychologist at three week intervals. Visits gradually withdrawn as progress occurred. Joint visits for complex cases.
Quine[19]	One-to-one meeting with parents at home to agree behavioural programme (two appointments). Details written up for parents. (Based on Hewitt[14]) Mainly positive bedtime routine and graded change. Tailored to individual needs.	Until parents were satisfied with the progress made. Range 5-30 weeks.	Weekly visits from health-visitor initially and then frequency agreed with parents. There was a follow-up appointment after three months.

3.4 Extinction

There were no studies of extinction found that were Level 3 or above on the Maryland Scale (Table 3) therefore the effect of extinction on the sleep problems of disabled children is classified as unknown. There were seven very small before and after studies; the number of participants ranged from three to 15. Most of the participants had learning disabilities. One used graduated extinction[24] and six used non-graduated extinction.[20,23,25-26] Generally, extinction was described as being used in conjunction with a positive bedtime routine. The studies of non-graduated extinction all used a similar approach (see Box 4).

Box 4: Non-graduated extinction

a) Establish a positive and regular bedtime routine
b) Settle child into bed
c) Say goodnight and leave the bedroom
d) Ignore child's protestations and do not re-enter the room (except in case of illness)
e) If the child comes out of their room, take the child immediately back to be with minimum interaction
f) When child sleeps through the night give them positive attention in the morning and explain why

The study of graduated extinction used different schedules for each of the families.[24] Parents started with waiting three and five minutes before entering their child's bedroom and responding to their crying or protests. The length of time gradually increased each night.

Most of the studies reported that the intervention was explained to parents in a single one-to-one session, though this was not always fully reported. This session was accompanied by daily telephone contact with parents at least on the days following initial implementation of extinction (Table 7). In one study parents received three training sessions[26] and in one they received two two-hour sessions[25] (see Appendix D for full details). Two studies explicitly focused on partner support strategies as part of the intervention given to parents.[25-26] The aim was to facilitate consistent parenting and to teach communication and problem-solving skills that help partners

assist and encourage each other in their parenting tasks. Three studies had a set duration of implementation: two[20] and seven weeks.[25-26] The remaining studies used a variable duration (see Table 8).

Table 6: Details of participants (extinction studies)

Study	Disability	Age	Baseline severity of sleep problem
Bramble[20] n=15	Severe LD	Mean 7.2yr Range 3.5-12yr	Severe sleep problem was an entry requirement. Mean severity 8 (SD 1.34) on 10-point VAS
Didden[21] n=3	Moderate LD; seizure disorder; mild LD with ADHD	Range 9.2-12.4yrs	Mean duration of night-time disruption ranged from 44min to 131min
Didden[22] n=4	Severe LD; moderate LD; mild LD	Range 1yr 11mth-25yr	Mean duration of night-time disruption ranged from 27min (SD 20.9) to 45min (SD 29.2)
Didden[23] n=6	Spinal muscle atrophy, ADHD, Prader-Willi syndrome†	Range 2-4yrs	Mean duration of night-time disruption ranged from 21 to 131min
Durand[24]	Mild to moderate LD, pervasive developmental delay, autism	Range 2 -12yr	% of nights with bedtime disturbance range from 65% to 100% and night waking from 36% to 94% of nights
Thackery[25] n=3	Severe LD; moderate LD; mild LD	Range 5-10yrs	Based on BEDS questionnaire had clinically significant sleep problems
Weiskop[26] n=13	Autism; Asperger syndrome; fragile x syndrome	Mean 5yrs Range 1yr 1mth-9yr 1mth	Unclear; Problems reported were bedtime disturbances, sleeping in parental bed, night waking and disruptive behaviour

VAS: visual analogue scale, BEDS: Behavioural Evaluation of Disorders of Sleep questionnaire, †The study included six children but one had sleep terrors and one had sleep problems related to epilepsy which were not relevant to the review. Before and after data were available for three of the remaining four children and data were extracted for these three only.

All of the studies reported improvement in children's sleep problems following the intervention though because of the study design it is unclear whether improvement can be directly attributed to the intervention. Three of the studies reported an extinction burst in some children (i.e. a temporary increase in severity of the target behaviour following the first days of implementation of the intervention): this occurred in seven out of 13 children,[26] two out of three;[25] and one out of four.[22] (See Appendix D for full details of the individual study results.)

One of the benefits put forward for use of extinction is that improved behaviour can occur over a shorter period of time than a graduated behavioural approach. From the information available in these studies there appears to be considerable variability in how rapid the response is. Only one study explicitly measured time to response. Bramble asked parents how long it took for their child to positively respond to the extinction technique. The mean time within which change was observed by parents was 3.6 nights (SD 1.9, range 1 to 7).[20] However, in the studies using a variable duration of intervention depending on response to treatment, the length of time is considerably longer (Table 8). This may be due to differences between the studies in factors such as the severity of the participants' sleep, the motivation of parents, how they were selected for the study, how rigorously parents implemented the intervention and/or the quality of the training they received.

Weiskop *et al.*[26] who conducted one of the two larger studies of extinction (13 participants), observed that extinction did not seem appropriate for early morning waking or night rocking possibly because they were not positively reinforced by parental responses. Two children who were withdrawn from their study were older and more non-compliant than those who remained: the authors suggest that extinction may be too difficult or stressful to implement with extremely non-compliant or older children.

Three studies formally elicited parents' views on extinction. One study, using the Program Evaluation Questionnaire (PEQ), reported that the best aspects of the programme were the good outcome, the support provided and the training, record-keeping was the aspect they liked least. Two parents reported that it was difficult to stick to a bedtime routine, one found the training sessions too long and three thought

the programme was too time-consuming.[26] Another study using the PEQ reported that the three parents were very satisfied with the outcomes of the intervention and the techniques used. They thought the programme was very appropriate for their child and would strongly recommend it to a friend. They particularly like the support they received but did not like ignoring their child when they called.[25] The third study, which was conducted in the UK, reported that in terms of the acceptability of the approach 12 parents thought the treatment approach was 'just right' for their child and three thought it was 'rather tough'. There was high overall satisfaction with the treatment amongst parents.[20] The authors of two studies commented that parents found the intervention difficult to implement, though were satisfied with the results.[21] In the study of graduated extinction the authors stated that parents were at first hesitant to delay attending to their children but found the short delay easy to tolerate.[24]

Table 7: Details of intervention (extinction studies)

Study	Details of intervention	Duration of implementation	Support for parents
Bramble[20]	Regular and positive bedtime routine. For extinction parents were instructed to rapidly settle child, leave bedroom, ignore child protestations unless in case of illness, if child leaves room after settling time firmly tell child to return to bed and, if necessary physically carry back to bed with minimal affective contact. Treatment was explained in single on-to-one session at home or clinic.	Two weeks	Telephone contact on the three days following the first session to offer encouragement and deal with problems. Additional telephone contact as needed. Only a minority required more than four calls.
Didden[21]	Extinction (similar to above)[†] There was at least one meeting with parents at home to conduct a functional assessment and provide information on the technique.	40 and 80 nights (approx six and 11 weeks)	Daily telephone contact. The authors state that this was an important part of the intervention especially during initial treatment.
Didden[22]	Extinction (similar to above) There was at least one meeting with parents at home to conduct a functional assessment and provide information on the technique.	10 to 120 nights	Not explicitly stated though the authors advise daily contact between parents and therapist especially in the first week of treatment.
Didden[23]	Extinction (similar to above).	29 to 54 nights	Not explicitly stated.
Durand[24]	Graduated extinction and consistent bedtime routine. The extinction schedule varied between children. In response to night waking or disruptive behaviour neutral and minimal reassurance was provided at gradually increasing intervals e.g. one parent started by waiting three minutes before entering the room and the delay was increased by two minutes each night. Two one-to-one meetings with parents.	8 to 16 weeks	Regular telephone contact during baseline and treatment sessions.

Study	Details of intervention	Duration of implementation	Support for parents
Thackeray[25]	Extinction with regular and positive bedtime routine, reinforcement, effective instructions and partner support. Two week training programme delivered individually to parents at clinic (based on McDonald & Patzold five Step Sleep Programme).	Seven weeks	Telephone contact on at least three of the mornings after extinction implemented and weekly during the rest of the programme. There was 6 hours face-to-face contact in total.
Weiskop[26]	Extinction with regular and positive bedtime routine, reinforcement, effective instructions and partner support. Initial interview and functional assessment followed by three, weekly training sessions delivered individually to parents (at home and clinic). The different types of extinction were explained. All parents chose standard extinction which was also the therapist's preference.	Seven weeks	Daily telephone contact in the days following implementation of extinction and weekly during the rest of the programme. There was also a review session after training ended.

† A single child received differential reinforcement of incompatible behaviours Details not reported as only single case)

3.5 Sleep restriction

There were no studies of sleep restriction found that were Level 3 or above on the Maryland Scale (Table 3) therefore the effect of sleep restriction on the sleep problems of disabled children is classified as unknown. There were two small studies of two[28] and four[27] participants where sleep restriction was used in conjunction with a positive bedtime routine. This intervention involved restricting the amount of time the child slept in bed to 90 per cent of the total time that the child normally slept at baseline. The child's bedtime and/or wake-time were adjusted for the new schedule. The intention is that this can be faded back to an age appropriate length of sleep time at the end of the intervention. Parents were also instructed to establish consistent bedtime routines (See Appendix D for full details). The extent of support received by parents in the two studies was unclear. Both studies reported improvements in child sleep problems (see Appendix D for full details of results) though because of the study design it is unclear whether improvement can be directly attributed to the intervention. One child experienced an increase in sleep-walking by the third week of the intervention (mean 2.3 episodes per week). This child also experienced two episodes of sleep terrors during the intervention.[28] The views of parents were not formally elicited. The authors of both studies stated that the parents found the intervention easy to implement on a regular basis. They suggest that the intervention is suitable for parents who are uncomfortable about using extinction or graduated extinction.

Table 8: Details of participants (sleep restriction studies)

Study	Disability	Age	Baseline severity of sleep problem
Christodulu[27] n=4	Developmental disabilities	Range 2yr 9mth to 5yr 11mth	Mean duration of bedtime disturbances ranged from 88 to 849 mins/week and duration of night waking from 92 to 682mins.
Durand[28] n=2	Autism; developmental delay	Both 4yr	Duration of bedtime disturbances 1.27hrs/week and 1.38 hrs/week.

Table 9: Details of intervention (sleep restriction studies)

Study	Details of intervention	Duration of implementation	Support for parents
Christodulu[27]	Sleep restriction and consistent bedtime routine. Positive bedtime routine was introduced first. Parents were also instructed to return children to their own bed if they got out of bed or got into parental bed. Sleep restriction involved restricting the amount of time the child was in bed to 90% of the time the child normally slept (based on parent sleep diaries). Bedtime and/or sleep time was adjusted for the new schedule.	Positive bedtime routine lasted from a few days to approximately six wks; sleep restriction plus bedtime routine lasted approximately 14 to 18wks	Details not provided.
Durand[28]	Sleep restriction and consistent bedtime routine. Consistent bedtime routines were established and parents were instructed to return children to their own bed if they got out of bed or got into parental bed. Sleep restriction involved restricting the amount of time the child was in bed to 90% of the time the child normally slept (based on parent sleep diaries). Bedtime and/or sleep time was adjusted for the new schedule.	Approximately 15 and 25 weeks	Details not provided.

3.6 Faded bedtime with response cost

There was one study of faded bedtime with response cost classified as above Level 3 on the Maryland Scale (Table 3). This was an RCT of 14 participants, using bedtime scheduling as a comparator, which had some methodological limitations (Appendix C).[29] There were also two before and after studies with three and four participants.[30-31] All of these studies were conducted in hospital settings in the US and it is unclear how easily such an intervention could be applied in the home setting. Full details of each of these studies are reported in Appendix D, though the intervention is not discussed in any detail here due to the lack of information on its use in a home-setting.

The intervention involved setting a bedtime at which sleep onset was highly likely within 15 minutes of being put to bed (this was half an hour later than the average time of sleep onset at baseline). A consistent bedtime routine was also established. The child was not permitted to go to sleep before the new bedtime and was woken at a set time each morning. The response cost occurred if the child did not fall asleep within 15 minutes: they were removed from bed and kept awake for one hour (played with toys, watched TV, etc.). They were then returned to bed and this was repeated until the child was put to bed and fell asleep within 15 minutes. If the child fell asleep within 15 minutes of bedtime, bedtime was made half an hour earlier the next night. If they did not fall asleep it was made half an hour later.

4. Discussion

4.1 Summary of the evidence

We conducted a rapid evidence review focusing on interventions for behavioural sleep problems in young disabled children (up to age eight years), specifically interventions that can be carried out by parents in the home. Of the 19 studies identified, four were RCTs and 15 were before and after studies, most of which had less than 10 participants. Three of the four RCTs had been conducted in a UK setting. The majority of participants had learning disabilities ranging from mild to severe and had serious sleep problems of long-standing duration.

Evidence was identified on three different behavioural approaches conducted in the home setting: interventions using multiple behavioural techniques (non-specific behavioural interventions); extinction (graduated and non-graduated); and sleep restriction. Evidence was also identified on faded bedtime with response cost; however this was implemented in an in-patient setting for most of the participants and it is unclear from the evidence available how easily this method would transfer to a home setting.

There were two types of non-specific behavioural interventions evaluated: general information giving and a more individually tailored intervention combining information giving to parents with an individual treatment plan for each child based on an assessment of the sleep problem. The main characteristic that these two groups of studies had in common was that they did not evaluate a single behavioural technique, but provided parents with information on a range of approaches. Two studies evaluated the provision of general information on behavioural techniques to parents, with the intention that parents would then implement the techniques with their children. There was evidence from a single RCT that a 90 minute session explaining behavioural approaches to child sleep problems, delivered to parents in their own home, was more effective than no intervention in reducing sleep disturbance post-treatment and six months later.[14] There was evidence from the same study that provision of the same information through a booklet only was also more effective than no intervention in reducing sleep disturbance over the same time

period. The second study (based on the main statistical analysis) did not find any benefit with a 90 minute instruction and discussion session with small groups of mothers.[15] Interpretation of this study is complicated by the inclusion of children with and without sleep problems.

Unfortunately there is not a large enough body of appropriate evidence to conclude that such an intervention works. Overall, there is sufficient evidence to conclude that the provision of information to parents of children with a severe learning disability and a severe behavioural sleep problem, either in a single face-to-face session or through a booklet, is a promising approach. Further research across a range of children with different disabilities is required. It would seem reasonable to conclude that such techniques would be transferable to other disabled groups. However, a key question is whether the parents who participated in the study are more highly motivated and/or feel more confident and are at a stage of readiness to deliver such an intervention with their children compared to a general population.

There were four studies that evaluated provision of information in conjunction with individual treatment plans. The interventions in these studies were more intensive than the two described above. In addition to the individual treatment plans parents were also provided with ongoing information and support (by telephone or face-to-face) while they implemented the techniques with their children. There was evidence from a single RCT of children with a severe learning disability and one or more daytime challenging behaviours.[16] A functional analysis of the individual children's sleep problem in combination with an agreed written behavioural programme delivered by parents and provision of information on behavioural techniques was more effective than no intervention in reducing sleep problems but not daytime challenging behaviour. There were also some benefits for parental outcomes in this study. The remaining three studies did not have a control group, though their findings supported the results from this RCT. Overall, there is evidence that the intervention in this RCT is a promising one for children with severe learning disabilities. One of the before and after studies also used a similar intervention in children with a range of chronic illnesses, as well as in children with learning disabilities.[17] Unfortunately outcome data were not reported for the two groups.

It is interesting that two interventions that vary intensity are both promising interventions.[14,16] Arguably in the study of the lower intensity interventions which focused on provision of information (either face-to-face or through a booklet) participants will probably have received support indirectly as researchers spent a total of 90 minutes with all participants gathering outcome data. Data was gathered from the control group in a similar way but it is possible that the contact in the two intervention groups encouraged parents to implement the intervention. But even if this was the case, the intervention was still less intensive in that there was not a functional assessment or a written action plan for each child.

It is unlikely that the lower intensity intervention is an appropriate approach for all families and some may prefer to have to a tailored intervention to implement rather than trying to apply general information to their own specific situation. Equally some parents may prefer to avoid the time commitment of a more intensive intervention. There would be benefit in evaluating the relative cost-effectiveness of the two approaches as well as parental preferences. In the absence of such information it may be beneficial, where practical, to make available the less intensive approach to all families in the first instance and to provide the more intensive approach to families who feel that they need the extra support or for whom the less intensive approach is not effective.

Although there were several studies evaluating extinction (mainly non-graduated extinction), no controlled studies were identified. In the absence of a control or comparison group there is uncertainty as to whether the improvement evident in the studies was a direct result of the intervention. However, given that sleeping problems in children with learning disabilities can be long-standing and unlikely to spontaneously improve (it was stated in several studies that parents had already tried other approaches that had failed) these studies indicate that extinction may be a feasible approach to use. An argument for the use of extinction is that improvement may be quicker than with other graduated methods. Based on the group of included studies there was considerable variability across and within studies in the length of time for benefit to occur. One of the disadvantages of extinction is that parents need to leave the child to cry if they do so after they are put to bed. This may be difficult for some parents to tolerate. Overall the parents in these small studies were positive

about the approach, though some expressed that they disliked ignoring their child. Parents may have been selected or selected themselves into these studies on the basis of their finding extinction acceptable therefore it is unclear how acceptable the technique would be to parents of disabled children general.

As with extinction, only before and after studies were available on sleep restriction. Both studies showed improvement in sleep outcomes but because of the study design it is unclear whether the improvement can be directly attributed to the intervention. The authors suggest that sleep restriction may be particularly suitable for parents who are uncomfortable about using extinction or graduated extinction.

4.2 Gaps in the evidence

Previous reviews in this field in 1999[5] and 2000[39] highlighted the need for further and better research. Some valuable work has been done since then, in particular in the UK; however, the evidence base remains limited. Further research is required on behavioural interventions for behavioural sleep problems in young children with disabilities; in particular there is a lack of studies with a control or comparison group. Ideally future studies would compare different types of interventions, though, as highlighted by Montgomery this may not be practical due to the large number of participants required. Further research on the longer-term outcomes following a behavioural intervention is also required. Do any short-term benefits continue into the long-term or do parents need refresher courses and/or longer term follow-up?

The interventions in the included studies are effectively complex multi-component interventions and it is unclear from the studies what aspects of the interventions are essential for a beneficial effect. For example, as pointed out by Hewitt,[18] in addition to the specific behavioural technique, factors such as directly involving parents, a written treatment programme, daily feedback for parents from diaries and weekly support visits may have been important.

The components that are important for an effective intervention may also vary depending on the particular needs of parents it is being delivered to. While there was evidence that a booklet alone was effective in one study, in another study one third of

parents needed a preliminary intervention before they were ready to cope with the main intervention. This emphasises the importance of being aware of the needs of parents as well as focusing on the behavioural sleep problems of the child. Most of the included studies did not formally elicit the views of parents therefore it is unclear what parents' views were about some of the approaches and what aspects of the interventions they found most helpful. Further research on this would be helpful in developing future services. In particular, a clearer perspective on parents' views, and on the views of professionals who provide interventions, is required in relation to 'real-life' services and interventions, as opposed to specifically within the context of a research study evaluating effectiveness. As Robinson and Richdale,[40] little is known about interventions offered to families in 'real-life' settings.

The participants in the studies did not cover the whole spectrum of children's disabilities. Most of the participants in the included studies had a range of learning disabilities from mild to severe. Further evidence is required on the issues around delivering such interventions to children with other disabilities and children with complex health needs, for example, children with physical conditions which require night-time assistance and medication in addition to a behavioural sleep problem.

Sleep problems may be both behavioural and physical and this points to the need for careful evaluation of disabled children's sleep problems before planning an intervention. Stores and Wiggs[41] suggest that a three tier service is needed:

- *Primary care*, for relatively straightforward sleep problems, for example settling or night waking problems, which can be treated by health visitors or GPs;
- Community or hospital *paediatric services* for more difficult diagnostic or treatment problems; and
- *Specialised sleep disorder services,* at a regional level, for the most complex problems.

For this system to be effective, all personnel involved need to have basic training in identifying and managing sleep disorders.

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Appendix A: Search Strategy

The search strategies used to search the databases are described in detail below.

Cochrane Database of Systematic Reviews (CDSR), DARE and CENTRAL

#1 MeSH descriptor Sleep Disorders explode all trees
#2 (sleep* or night* or nocturnal):ti,ab,kw
#3 (bedtime or "bed time" or settl* or waking or wake*):ti,ab,kw
#4 (#1 OR #2 OR #3)
#5 (infant* or baby or babies or toddler* or child* or preschool*):ti,ab,kw
#6 MeSH descriptor Disabled Persons explode all trees
#7 MeSH descriptor Mental Disorders explode all trees
#8 (disabled or disability or disabilities or handicap* or retard* or autist* or asperger* or blind or blindness or deaf or deafness or or (attention near/2 deficit) or adhd):ti,ab,kw
#9 (intellectual* impair*):ti,ab,kw
#10 ("complex needs" or "special needs"):ti,ab,kw
#11 ((life near limit*) or (life near threaten*)):ti,ab,kw
#12 (learning near (disorder* or disab*)):ti,ab,kw
#13 (technolog* near depend*):ti,ab,kw
#14 ((cerebral palsy) or ("down* NEAR/2 syndrome")):ti,ab,kw
#15 (#6 OR #7 OR #8 OR #9 OR #10 OR #11 OR #12 OR #13 OR #14)
#16 MeSH descriptor Psychotherapy explode all trees
#17 (behav* near (intervention* or therap* or treatment* or program* or approach* or techniqu* or strateg*)):ti,ab,kw
#18 (avers* near/2 therap*):ti,ab,kw
#19 (biofeedback or chronotherap* or (contingency next manage*) or extinction or (negative next consequence*) or schedul*):ti,ab,kw
#20 (reinforc* or routine* or (response next cost*) or separation or desensit* or (omission next train*) or faded or fading):ti,ab,kw
#21 (cbt or (cognitive near/3 therap*)):ti,ab,kw
#22 (#16 OR #17 OR #18 OR #19 OR #20 OR #21)
#23 (#4 AND #5 AND #15 AND #22)

MEDLINE, Ovid MEDLINE(R) In-Process & Other Non-Indexed Citations and Ovid MEDLINE(R) <1950 to Present>

1 exp sleep disorders/ (41103)
2 ((sleep$ or night$ or nocturnal) adj3 (disturb$ or problem$ or behav$ or disorder$ or disrupt$ or difficult$ or regulat$ or habit$ or questionnaire$)).ti,ab. (23448)
3 (bedtime or bed time or settl$4 or sleepless$ or waking or wake$1 or wakeful$).ti,ab. (29939)
4 or/1-3 (75199)
5 exp child/ or exp infant/ (1684476)
6 exp child behavior/ or exp infant behavior/ (10514)
7 (infant$ or baby or babies or toddler$ or child or children or preschool$).ti,ab. (861375)
8 or/5-7 (1858948)
9 exp disabled persons/ (35898)
10 exp mental disorders diagnosed in childhood/ (112868)
11 (disabled or disability or disabilities or handicap$ or retard$).ti,ab. (168251)
12 intellectual$ impair$.ti,ab. (919)
13 ((complex or special) adj3 needs).ti,ab. (4372)
14 (life adj (limit$ or threaten$)).ti,ab. (35724)

15 learning disorder$.ti,ab. (676)
16 technolog$ depend$.ti,ab. (208)
17 (cerebral palsy or down$2 syndrome).ti,ab. (24456)
18 (autist$ or asperger$ or blind or blindness or deaf or deafness or adhd or attention deficit).ti,ab. (162781)
19 or/9-18 (464869)
20 exp psychotherapy/ (120601)
21 (behav$ adj3 (intervention$ or therap$ or treatment$ or program$ or approach$ or techniqu$ or strateg$)).ti,ab. (28975)
22 avers$ therap$.ti,ab. (202)
23 (biofeedback or chronotherap$ or contingency manage$ or extinction or negative consequence$ or schedul$).ti,ab. (93784)
24 (reinforc$ or routine$ or response cost$ or separation or desensit$ or omission train$ or faded or fading).ti,ab. (352585)
25 (cbt or (cognitive adj3 therap$)).ti,ab. (6930)
26 or/20-25 (566280)
27 4 and 8 and 19 and 26 (335)
28 limit 27 to (english language and yr="1985 - 2008") (260)
29 limit 28 to (case reports or comment or editorial or letter) (39)
30 28 not 29 (221)

The search was amended on 23/9/8 to search for 'delayed development' by adding in an additional search line as follows to disability concept:

(develop$ adj3 delay$).ti,ab.

Three new records were identified from MEDLINE, but all had already been found from other searches.

EMBASE, OvidSP, <980 to 2008 Week 33>

1 exp sleep disorders/ (70163)
2 ((sleep$ or night$ or nocturnal) adj3 (disturb$ or problem$ or behav$ or disorder$ or disrupt$ or difficult$ or regulat$ or habit$ or questionnaire$)).ti,ab. (19942)
3 (bedtime or bed time or settl$4 or sleepless$ or waking or wake$1 or wakeful$).ti,ab. (24838)
4 or/1-3 (95487)
5 exp child behavior/ or exp infant behavior/ (12472)
6 (infant$ or baby or babies or toddler$ or child or children or preschool$).ti,ab. (545840)
7 limit 4 to (infant <to one year> or child <unspecified age> or preschool child <1 to 6 years> or school child <7 to 12 years>) (8686)
8 (4 and (5 or 6)) or 7 (11644)
9 exp Disabled Person/ (2582)
10 exp Mental Disease/ (684814)
11 exp Disability/ (40748)
12 exp Handicapped Child/ (2719)
13 (disabled or disability or disabilities or handicap$ or retard$).ti,ab. (124793)
14 intellectual$ impair$.ti,ab. (776)
15 ((complex or special) adj3 needs).ti,ab. (2691)
16 (life adj (limit$ or threaten$)).ti,ab. (30533)
17 learning disorder$.ti,ab. (500)
18 technolog$ depend$.ti,ab. (126)
19 (cerebral palsy or down$2 syndrome).ti,ab. (18087)
20 (autist$ or asperger$ or blind or blindness or deaf or deafness or adhd or attention deficit).ti,ab. (133842)
21 or/9-20 (917539)

22 exp psychotherapy/ (75871)
23 (behav$ adj3 (intervention$ or therap$ or treatment$ or program$ or approach$ or techniqu$ or strateg$)).ti,ab. (25578)
24 avers$ therap$.ti,ab. (112)
25 (biofeedback or chronotherap$ or contingency manage$ or extinction or negative consequence$ or schedul$).ti,ab. (73647)
26 (reinforc$ or routine$ or response cost$ or separation or desensit$ or omission train$ or faded or fading).ti,ab. (281295)
27 (cbt or (cognitive adj3 therap$)).ti,ab. (7750)
28 or/22-27 (431281)
29 8 and 28 and 21 (915)
30 limit 29 to (english language and yr="1985 - 2008") (814)
31 limit 30 to (editorial or letter or note) (21)
32 30 not 31 (793)

PsycINFO, OvidSP, <1967 to July Week 5 2008>
1 exp sleep apnea/ or exp sleep deprivation/ or exp sleep disorders/ or exp sleep onset/ or exp sleep talking/ or exp sleep treatment/ or exp sleep wake cycle/ or exp sleepiness/ or exp sleepwalking/ (11597)
2 ((sleep$ or night$ or nocturnal) adj3 (disturb$ or problem$ or behav$ or disorder$ or disrupt$ or difficult$ or regulat$ or habit$ or questionnaire$)).ti,ab. (10750)
3 (bedtime or bed time or settl$4 or sleepless$ or waking or wake$1 or wakeful$).ti,ab. (12812)
4 or/1-3 (26111)
5 limit 4 to 100 childhood <birth to age 12 yrs> (3038)
6 exp childhood development/ (44795)
7 (infant$ or baby or babies or toddler$ or child or children or preschool$).ti,ab. (355589)
8 (4 and (6 or 7)) or 5 (4024)
9 exp disabilities/ (38564)
10 exp mental disorders/ (315804)
11 exp mental retardation/ (34781)
12 exp learning disorders/ (25979)
13 exp attention deficit disorder/ (12050)
14 (disabled or disability or disabilities or handicap$ or retard$).ti,ab. (103746)
15 intellectual$ impair$.ti,ab. (790)
16 ((complex or special) adj3 needs).ti,ab. (5099)
17 (life adj (limit$ or threaten$)).ti,ab. (2695)
18 learning disorder$.ti,ab. (971)
19 technolog$ depend$.ti,ab. (61)
20 (cerebral palsy or down$2 syndrome).ti,ab. (7027)
21 (autist$ or asperger$ or blind or blindness or deaf or deafness or adhd or attention deficit).ti,ab. (52668)
22 or/9-21 (456767)
23 exp behavior modification/ (34956)
24 exp psychotherapy/ (142119)
25 (behav$ adj3 (intervention$ or therap$ or treatment$ or program$ or approach$ or techniqu$ or strateg$)).ti,ab. (50670)
26 avers$ therap$.ti,ab. (357)
27 (biofeedback or chronotherap$ or contingency manage$ or extinction or negative consequence$ or schedul$).ti,ab. (46417)
28 (reinforc$ or routine$ or response cost$ or separation or desensit$ or omission train$ or faded or fading).ti,ab. (85220)
29 (cbt or (cognitive adj3 therap$)).ti,ab. (13138)
30 or/23-29 (291706)
31 8 and 22 and 30 (274)

32 limit 31 to (english language and yr="1985 - 2008") (226)
33 limit 32 to ("comment/reply" or editorial or letter) (4)
34 32 not 33 (222)
35 from 34 keep 1-222 (222)

The search was amended on 23/9/8 to search for 'delayed development' by adding in an additional search line, as follows, to disability concept:

(develop$ adj3 delay$).ti,ab.

Five new records were identified from PsyCINFO. Three of these had already been found from other searches.

CINAHL, OvidSP, <1982 to August Week 3 2008>
1 exp sleep disorders/ (7241)
2 ((sleep$ or night$ or nocturnal) adj3 (disturb$ or problem$ or behav$ or disorder$ or disrupt$ or difficult$ or regulat$ or habit$ or questionnaire$)).ti,ab. (3325)
3 (bedtime or bed time or settl$4 or sleepless$ or waking or wake$1 or wakeful$).ti,ab. (2889)
4 or/1-3 (10802)
5 exp child/ or exp infant/ (170003)
6 exp child behavior/ or exp infant behavior/ (3656)
7 (infant$ or baby or babies or toddler$ or child or children or preschool$).ti,ab. (105377)
8 or/5-7 (192886)
9 exp disabled/ (16225)
10 exp mental disorders/ (124183)
11 exp developmental disabilities/ (2156)
12 (disabled or disability or disabilities or handicap$ or retard$).ti,ab. (30208)
13 intellectual$ impair$.ti,ab. (99)
14 ((complex or special) adj3 needs).ti,ab. (2765)
15 (life adj (limit$ or threaten$)).ti,ab. (4246)
16 learning disorder$.ti,ab. (82)
17 technolog$ depend$.ti,ab. (134)
18 (cerebral palsy or down$2 syndrome).ti,ab. (3693)
19 (autist$ or asperger$ or blind or blindness or deaf or deafness or adhd or attention deficit).ti,ab. (15261)
20 or/9-19 (170487)
21 exp psychotherapy/ (47175)
22 (behav$ adj3 (intervention$ or therap$ or treatment$ or program$ or approach$ or techniqu$ or strateg$)).ti,ab. (6229)
23 avers$ therap$.ti,ab. (7)
24 (biofeedback or chronotherap$ or contingency manage$ or extinction or negative consequence$ or schedul$).ti,ab. (8557)
25 (reinforc$ or routine$ or response cost$ or separation or desensit$ or omission train$ or faded or fading).ti,ab. (20842)
26 (cbt or (cognitive adj3 therap$)).ti,ab. (1825)
27 or/21-26 (77320)
28 4 and 8 and 20 and 27 (72)
29 limit 28 to (english language and yr="1985 - 2008") (69)

SPECTR and C2-RIPE (Campbell Collaboration), http://geb9101.gse.upenn.edu
(sleep) or (wake) or (waking) or (night) or (bedtime) or ("bed time") (in either "indexed" or "non-inedxed" fields)
AND

(infant) or (baby) or (babies) or (toddler) or (child) or (preschool) (in either "indexed" or "non-indexed" fields)

HMIC, OvidSP, < July 2008 >

1 sleep$.mp. (526)
2 ((sleep$ or night$ or nocturnal) adj3 (disturb$ or problem$ or behav$ or disorder$ or disrupt$ or difficult$ or regulat$ or habit$ or questionnaire$)).mp. (221)
3 (bedtime or bed time or settl$4 or sleepless$ or waking or wake$1 or wakeful$).mp. (481)
4 exp sleep/ or exp sleep disorders/ (130)
5 or/1-4 (1024)
6 child$.mp. or exp children/ (24726)
7 (infant$ or baby or babies or toddler$ or preschool).mp. (3616)
8 or/6-7 (26362)
9 exp disabilities/ (27219)
10 (disabled or disabilit$ or handicap$ or retard$).mp. (14077)
11 (intellect$ adj2 impair$).mp. (23)
12 ((complex or special) adj3 needs).mp. (1013)
13 (life adj (limit$ or threaten$)).mp. (299)
14 technolog$ depend$.mp. (14)
15 (cerebral palsy or down$2 syndrome).mp. (314)
16 (autist$ or asperger$ or blind or blindness or deaf or deafness or adhd or attention deficit).mp. (1393)
17 (learning adj3 (disab$ or disorder$)).mp. (5570)
18 or/9-17 (31892)
19 exp psychotherapy/ (1946)
20 (behav$ adj3 (intervention$ or therap$ or treatment$ or program$ or approach$ or techniqu$ or strateg$)).mp. (1083)
21 avers$ therap$.mp. (3)
22 (biofeedback or chronotherap$ or contingency manage$ or extinction or negative consequence$ or schedul$).mp. (1419)
23 (reinforc$ or routine$ or response cost$ or separation or desensit$ or omission train$ or faded or fading).mp. (4832)
24 (cbt or (cognitive adj3 therap$)).mp. (229)
25 or/19-24 (8890)
26 25 and 8 and 18 and 5 (12)
27 limit 26 to yr="1985 - 2010" (11)

NNR archive, https://portal.nihr.ac.uk/Pages/NRRArchiveSearch.aspx.

This is a difficult interface to search. Searches have to be constructed with the most general concept first and then more specific concepts used to narrow down the retrieved set. There is no facility to record the search history or to export the results.

"sleep*" or "wake*" or waking or bedtime or "settl*" or "night*"
AND
"infan*" or baby or babies or "toddler*" or "child*" or "preschool*"
AND
"disab*" or "disorder*" or "handicap*" or "retard*" or "impair*" or special or palsy or syndrome or "autis*" or "asperger*" or "blind*" or "deaf*" or adhd

sleep*
AND
child OR infant
AND

psychotherapy OR behavior-therapy OR “cognitive*” OR biofeedback

CERUK, http://www.ceruk.ac.uk/

Search terms were entered one by one.

Sleep*
Waking
Wake*
bedtime
“bed time”
Night*
settl*

ERIC, Dialog/Datastar
sleep OR bedtime OR bed ADJ time OR settl$4 OR sleepless$ OR waking OR wake$1 OR wakeful$ OR ((sleep$ OR night$ OR nocturnal) NEAR (disturb$ OR problem$ OR behav$ OR disorder$ OR disrupt$ OR difficult$ OR regulat$ OR habit$ OR questionnaire$))
AND
(Children#.W..DE.) OR (Child-Behavior#.W..DE.) OR (infant$ OR baby OR babies OR toddler$ OR child OR children OR preschool$)
AND
(Disabilities#.W..DE.) OR (disabled OR disability OR disabilities OR handicap$ OR retard$) OR (intellectual$ NEAR impair$) OR ((complex OR special) NEAR needs) OR (life ADJ (limit$ OR threaten$)) OR (learning ADJ (disorder$ OR disab$)) OR (technolog$ ADJ depend$) OR (cerebral ADJ palsy OR down$2 ADJ syndrome OR autist$ OR asperger$ OR blind OR blindness OR deaf OR deafness OR adhd OR attention ADJ deficit))
AND
(Conditioning#.W..DE.) OR (Psychotherapy#.W..DE.) OR (behav$ NEAR (intervention$ OR therap$ OR treatment$ OR program$ OR approach$ OR techniqu$ OR strateg$)) OR (avers$ ADJ therap$) OR biofeedback OR chronotherap$ OR contingency ADJ manage$ OR extinction OR negative ADJ consequence$ OR schedul$ OR reinforc$ OR routine$ OR response ADJ cost$ OR separation OR desensit$ OR omission ADJ train$ OR faded OR fading OR (cbt OR cognitive NEAR therap$)

limited to English language and publication date 1985 or later

Childdata
The search interface does not allow complex searches so a series of searches was undertaken:
sleep OR bedtime OR bed OR settling OR sleepless OR sleeplessness OR waking OR wakeful
sleep/title and disability/keyword
Sleep/abstract and disability/keyword
bed/title and disability/keyword
bed/abstract and disability/keyword
settling/title and disability/keyword
settling/abstract and disability/keyword
sleepless/title and disability/keyword
sleepless/abstract and disability/keyword
sleeplessness/title and disability/keyword
sleeplessness/abstract and disability/keyword
wakeful/title and disability/keyword

wakeful/abstract and disability/keyword

British Education Index, Dialog/Datastar, 1975 to date (BREI) and Australian Education Index

These databases were searched together and the results downloaded together

1 sleep.DE. 24
2 sleep.TI,AB. 26
3 (bed ADJ time).TI,AB. 0
4 bedtime.TI,AB. 2
5 settl$.TI,AB. 52
6 (sleepless$ OR waking OR wake$1 OR wakeful$).TI,AB. 26
7 sleeplessness 2
8 waking 4
9 (disturb$ OR problem$ OR behav$ OR disorder$ OR disrupt$ OR difficult$ OR regulat$ OR habit$ OR questionnaire$).TI,AB. 9034
10 (1 OR 2 OR 3 OR 4 OR 5 OR 6 OR 7 OR 8) AND 9 15
11 children 20041
12 PRIMARY-SCHOOL-STUDENTS.DE. OR CHILDREN#.W..DE. 8124
13 CHILD-BEHAVIOUR#.DE. 0
14 (infant$ OR baby OR babies OR toddler$ OR child OR children OR preschool$).TI,AB. 12989
15 students 20276
16 students 20276
17 ages 2117
18 11 OR 12 OR 13 OR 14 20412
19 10 AND 18 10
20 DISABILITIES#.W..DE. OR SPECIAL-NEEDS-STUDENTS.DE. OR MENTAL-RETARDATION.DE. OR READING-DIFFICULTIES.DE. OR AUTISM.W..DE. 8076
21 disabled OR disability OR disabilities OR handicap$ OR retard$ OR intellectual$ NEAR impair$ OR (complex OR special) NEAR needs OR life ADJ (limit$ OR threaten$) OR learning ADJ (disorder$ OR disab$) OR technolog$ ADJ depend$ OR (cerebral ADJ palsy OR down$2 ADJ syndrome OR autist$ OR asperger$ OR blind OR blindness OR deaf OR deafness OR adhd OR attention ADJ deficit).TI,AB. 9333
22 19 AND (20 OR 21) 7

Appendix B: Excluded Studies (from full paper screening)

Adlington, K., A. J. Liu, and R. Nanan. 2006. "Sleep disturbances in the disabled child--a case report and literature review." *Australian Family Physician* 35:711-715.	Not a primary study (review/discussion paper)
Bartlet, L. B. 2006. "Treating the sleep disorders of childhood: Current practice in the United Kingdom." *Journal of Indian Association for Child and Adolescent Mental Health* 2:89-95.	Not a primary study (review/discussion paper)
Didden, R., P.C. Duker, and H. Korzilius. 1997. "Meta-analytic study on treatment effectiveness for problem behaviours with individuals who have mental retardation." *American Journal on Mental Retardation* 101:387-399.	Not a primary study
Buschbacher, Pamelazita, Lise Fox, and Shelley Clarke. 2004. "Recapturing Desired Family Routines: A Parent-Professional Behavioral Collaboration." Pp. 15-39, *Research and Practice for Persons with Severe Disabilities RPSD*.	Case study; no sleep problem
Dorris, Liam, Nicola Scott, Sameer Zuberi, Neil Gibson, and Colin Espie. 2008. "Sleep problems in children with neurological disorders." *Developmental neurorehabilitation* 11:95-114.	Not a primary study (review/discussion paper)
Espie, C. A., and A. Wilson. 1993. "Improving sleep-wake schedules amongst people with mental handicaps: Some preliminary case material." *Behavioural Psychotherapy* 21:51-55.	None of the participants were under 8 years old
France, K. G., J. M. T. Henderson, and S. M. Hudson. 1996. "Fact, act, and tact: A three-stage approach to treating the sleep problems of infants and young children." *Child and Adolescent Psychiatric Clinics of North America* 5:581-599.	Not a primary study (review/discussion paper)
Glaze, D. G., C. L. Rosen, and J. A. Owens. 2002. "Toward a practical definition of pediatric insomnia." *Current Therapeutic Research - Clinical and Experimental* 63:B4-B17.	Not a primary study (review/discussion paper)
Hoban, T. F. 2000. "Sleeplessness in children with neurodevelopmental disorders: Epidemiology and management." *CNS Drugs* 14:11-22.	Not a primary study (review/discussion paper)

Johnson, K. P., and B. A. Malow. 2008. "Sleep in children with autism spectrum disorders." *Current Neurology and Neuroscience Reports* 8:155-161.	Not a primary study (review/discussion paper)
Johnson, Cynthia R. 1996. "Sleep Problems in Children with Mental Retardation and Autism." *Child and Adolescent Psychiatric Clinics of North America* 5:673-683.	Not a primary study (review/discussion paper)
Keenan, Ruth A., Matt R. Wild, Irene McArthur, and Colin A. Espie. 2007. "Children with developmental disabilities and sleep problems: Parental beliefs and treatment acceptability." *Journal of Applied Research in Intellectual Disabilities* 20:455-465.	Does not evaluate an intervention (survey of parents)
Krakowiak, Paula, Beth Goodlin-Jones, Irva Hertz-Picciotto, Lisa A. Croen, and Robin L. Hansen. 2008. "Sleep problems in children with autism spectrum disorders, developmental delays, and typical development: a population-based study." *Journal of Sleep Research* 17:197-206.	No intervention (prevalence study)
Lancioni, Giulio E., Reilly Mark F. O, and Gabriella Basili. 1999. "Review of Strategies for Treating Sleep Problems in Persons with Severe or Profound Mental Retardation or Multiple Handicaps." *American Journal on Mental Retardation* 104:170-186.	Not a primary study (review/discussion paper)
Lucas, P., K. Liabo, and H. Roberts. 2002. "Do behavioural treatments for sleep disorders in children with Down's syndrome work?" *Archives of Disease in Childhood* 87:413-414.	Review of reviews
Meltzer, Lisa J., and Jodi A. Mindell. 2004. "Nonpharmacologic treatments for pediatric sleeplessness." *Pediatric Clinics of North America* 51:135-151.	Not a primary study (review/discussion paper)
Morgenthaler, T. I., *et al.* 2006. "Practice parameters for behavioral treatment of bedtime problems and night wakings in infants and young children." *Sleep* 29:1277-1281.	Not a primary study (Report of American Academy of Sleep Medicine)
Morris, S., I. S. James-Roberts, J. Sleep, and P. Gillham. 2001. "Economic evaluation of strategies for managing crying and sleeping problems." *Archives of Disease in Childhood* 84:15-19.	Not disabled children

O'Callaghan, F. J., A. A. Clarke, E. Hancock, A. Hunt, and J. P. Osborne. 1999. "Use of melatonin to treat sleep disorders in tuberous sclerosis." *Developmental Medicine and Child Neurology* 41:123-126.	Not a behavioural intervention
Okawa, M., T. Nanami, S. Wada, T. Shimizu, and *et al.* 1987. "Four congenitally blind children with circadian sleep-wake rhythm disorder." *Sleep: Journal of Sleep Research & Sleep Medicine* 10:101-110.	Not a behavioural intervention
Paavonen, E., Taina Nieminen-von Wendt, Raija Vanhala, Eeva T. Aronen, and Lennart von Wendt. 2003. "Effectiveness of melatonin in the treatment of sleep disturbances in children with Asperger disorder." *Journal of Child and Adolescent Psychopharmacology* 13:83-95.	Not a behavioural intervention
Piazza, Cathleen C., and Wayne W. Fisher. 1991. "Bedtime fading in the treatment of pediatric insomnia." *Journal of Behavior Therapy and Experimental Psychiatry* 22:53-56.	Single case with disability as defined for purposes of the project
Quine, L. 1991. "Sleep problems in children with mental handicap." *Journal of Mental Deficiency Research* 35:269-290.	No intervention (prevalence study)
Richdale, Amanda L. 1999. "Sleep problems in autism: Prevalence, cause and intervention." *Developmental Medicine and Child Neurology* 41:60-66.	Not a primary study (review/discussion paper)
Roane, Henry S., Cathleen C. Piazza, Laura E. Bodnar, and Kerri L. Zimmerman. 2000. "Sleep Difficulties in Children with Developmental Disabilities." *Infants and Young Children* 13:1-8.	Not a primary study (review/discussion paper)
Robinson, A., and A. Richdale. 2004. "Sleep problems in children with an intellectual disability: Parental perceptions of sleep problems, and views of treatment effectiveness." *Child: Care, Health and Development* 30:139-150.	No intervention (survey)
Schreck, K. A. 2001. "Behavioral treatments for sleep problems in autism: Empirically supported or just universally accepted?" *Behavioral Interventions* 16:265-278.	Not a primary study (review/discussion paper)
Stores, Gregory. 1992. "Sleep studies in children with a	Not a primary study

mental handicap." *Journal of Child Psychology and Psychiatry* 33:1303-1317.	(review/discussion paper)
Stores, G., and L. Wiggs. 2001. *Sleep disturbance in children and adolescents with disorders of development: its significance and management*. London: Mac Keith Press.	No primary studies not already identified
Stores, G. 2001. *A clinical guide to sleep disorders in children and adolescents*. Cambridge: Cambridge University Press.	No primary studies not already identified
Turk, J. 2003. "Melatonin supplementation for severe and intractable sleep disturbance in young people with genetically determined developmental disabilities: short review and commentary." *Journal of Medical Genetics* 40:793-796.	Not a primary study (review/discussion paper)
Wiggs, L., G. Stores. 2006. “A randomised controlled trial of behavioural intervention for sleeplessness in children with autism spectrum disorders.” Journal of Sleep Research 15 (Suppl 1): S83	Only available as an abstract
Wiggs, L., and K. France. 2000. "Behavioural treatments for sleep problems in children and adolescents with physical illness, psychological problems or intellectual disabilities." *Sleep Medicine Reviews* 4:299-314.	Not a primary study (review/discussion paper)
Wiggs, L., and G. Stores. 1996. "Sleep problems in children with severe intellectual disabilities: What help is being provided?" *Journal of Applied Research in Intellectual Disabilities* 9:160-165.	No intervention (survey of parents)

Appendix C: Quality Assessment of RCTs

	Montgomery[14]	Piazza[29]	Stores[15]	Wiggs[16]
A) SELECTION BIAS				
Are the individuals selected to participate likely to be representative of the target population?	Somewhat likely	Not likely	Somewhat likely	Not likely
What percentage of selected individuals agreed to participate?	75%	Unclear	60%	61%
Rate this section	Moderate	Weak	Moderate	Weak
B) STUDY DESIGN				
Was the study described as randomised?	Yes	Yes	Yes	Yes
If Yes, was the method described?	Yes	No	Yes	No
If Yes, was the method appropriate?	Yes	-	No	-
Rate this section	Strong	Weak	Moderate	Weak
C) CONFOUNDERS				
Were there important differences between groups prior to the intervention?	No	No[2]	No	No
If yes, indicate the percentage of relevant confounders that were controlled in the design or analysis?	N/A	N/A	N/A	N/A
Rate this section	Strong	Strong	Strong	Strong
E) DATA COLLECTION METHODS				
Were data collection tools shown to be valid?	Yes[1]	No	No	Yes
Were data collection tools shown to be reliable?	Yes[1]	Partial[3]	No	Yes
Rate this section	Strong	Moderate	Weak	Strong
F) WITHDRAWALS AND DROPOUTS				
Were withdrawals and dropouts reported in terms of numbers and/or reasons per group?	Yes	Yes	No	Yes
Indicate the percentage of participants completing the study	97%	100%	Unclear	97%
Rate this section	Strong	Strong	Unclear	Strong
H) ANALYSES				
Are the statistical methods appropriate for the study design?	Yes	Unclear[4]	Partial	Unclear
Is the analysis on an intention to treat basis?	Yes (at post-treatment)	Yes	Unclear	Unclear

[1] Based on statement by authors; [2] Hours of disturbed sleep at baseline seemed similar for both groups. Baseline disturbed sleep was used as a covariate in the analysis and this was statistically significant; [3] interobserver reliability; [4] unclear whether use of parametric appropriate.

Appendix D: Data Extraction

Publication details		
Author: Bartlet[17]	**Year:** 1998	**Related publications:**
Stated aim: To gain experience in treating the sleep disorders of children with disabilities and illness and to support their families.		
Study design: Before and after		
The participants		
Number: N=61	**Age:** Mean 4yrs, 11mths (range 11mths to 17yrs)	**Sex:** 40 male, 21 female
Type of disability: 22 with chronic illness (most commonly asthma and upper respiratory tract infections and ear problems); 39 with a disability (most commonly non-specific severe learning disability, severe learning disability and a co-morbid condition and autism).		
Sleep problem: 80% (n=49) with settling problems; 97% (n=59) with night-waking problems. 38% (n=23) had parasomnias. In 42% of families (n=26) parents stayed in the child's bed and in 74% of families the child stayed in the parents' bed occasionally or regularly. **How the sleeping problem was assessed:** The Southampton Sleep Management Schedule was used. Conducted by a psychiatrist and/or health visitor and took 1.5-2hrs. **Other information:** 67 children were referred to the project over one year, 61 took up assessment and 57 received treatment (4 moved away after initial contact).		
The intervention		
Setting: Home-based. This was a one year project located at Southampton General Hospital. It was staffed by a part-time experienced health visitor, a child psychiatrist 4hrs per week. **Type of behavioural intervention:** Cueing, graded change, extinction and positive reinforcement depending on the sleep problem and parental preferences. In a 'high proportion' of cases the intervention was based on graded change. **Description of intervention:** Details were not provided of the specific behavioural methods. Eight children were prescribed hypnotics for 2-3 weeks where there was frequent night-time wakening in the presence of parental fatigue. **Duration:** Treatment was discontinued when parents were satisfied with the progress made. **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face-to-face, telephone, booklet):** Following assessment families had one or two appointments with the project workers at home or at the hospital. Following this contact was usually by telephone. The mean number of calls was 4.95 and duration ranged from 5 to 60 minutes. Sleep diaries were used to plan and monitor progress. A *preliminary intervention* was required for many parents prior to being trained in the behavioural intervention. It was established early in the project that about one-third of parents of parents were not ready to become involved in a behavioural programme. Particular issues included physical exhaustion, disagreement between partners about the way forward, low self esteem, and a concern that the child would suffer as a result of the intervention. Tearfulness and feelings of hopelessness were common and three mothers were identified as clinically depressed and were referred to their GP for help. The aim of the preliminary intervention was to allow parents time to develop trusting relationships with the project workers and to give them time to contemplate changing their routines. Specific details were not provided other than that a holistic, dynamic approach was used with strategies such as understanding, support, empowerment and opportunities to talk through past traumatic experiences.		

Description of comparator: No comparator

The outcomes measures

Outcome 1: Sleep Disturbance Index (SDI)

Details of measurement: Eight-point scale developed by Quine (1991). Four factors (settling, night waking, parents up at night, child in parental bed) are each rated as being a problem less than twice per week (0), a problem 2-4 times per week (1) or more then 4 times per week (2). The minimum score is 0 and the maximum 8. Internal reliability is high (Cronbach's alpha = 0.78).

Outcome 2: Parent view of impact of intervention on sleep problem

Details of measurement: Parents were asked if the sleep disturbance was 'better', 'same' or 'worse' following the intervention.

Outcome 3: General Health Questionnaire (GHQ)-30

Details of measurement: Administered to mothers at assessment and follow-up by postal questionnaire. Scores above 4/5 defined as 'high' and associated in many cases with psychological distress. Sensitivity 74%, Specificity 82%.

Length of follow-up: 3 to 6 months after end of treatment

Summary of the results:

- SDI (n=57) – The mean score reduced from 6.36 at baseline to 2.81 at follow-up and this was statistically significant based on a one sample t-test (mean difference 3.544 (SD 3.57), p=0.0000)
- Parent view – 45 families said the sleep disturbance was 'better'; 10 said it was the 'same'; 2 said it was 'worse'.
- GHQ-30 (n=52) – Mean score at baseline was 10.90 (SD 3.93) and 61% (n=36) were in the 'high' category. There was a statistically significant improvement in the GHQ-30 score at follow-up (mean difference 4.308 (SD 5.31), p=0.00)

Any negative consequences: Two families thought the sleep problem was worse following the intervention.

Views of parents: The authors state that few parents opted for the extinction technique. Parents in seven families found the programmes difficult to manage or ineffective. The authors state that of the 27 parents who commented on the project, the tone of the remarks was that specialist help was useful and should be more readily available.

Authors' conclusion: Forty-five children improved as a result of the intervention but treatment was found to be more onerous than the literature suggests.

Publication details		
Author: Bramble[20]	**Year:** 1996	**Related publications:** Bramble[38]
Stated aim: To investigate the acceptability and safety of a behavioural modification programme aimed at the rapid extinction of night settling and night waking problems in children.		
Study design: Before and after		
The participants		
Number: N=15	**Age:** Mean 7.2yrs (range 3.5 to 12)	**Sex:** 10 male, 5 female
Type of disability: Severe learning disability (four children also had cerebral palsy and 3 had epilepsy)		
Sleep problem: Lifelong severe night settling and/or night waking **How the sleeping problem was assessed:** Severe problem was defined as the child taking at least an hour to settle at bedtime and waking up most nights and disturbing parents. **Other information:** The participants were taken from a continuous series of referrals to the clinic. The majority were referred by specialist community nurses, paediatricians or a child psychiatrist.		
The intervention		
Setting: Home-based **Type of behavioural intervention:** Extinction **Description of intervention:** Parents were given the following advice (based on Pearce 1991): 1) regular bedtime; 2) establish regular routine before bedtime and calm children down; 3) set mood for sleep rather than wakefulness and play before bedtime; 4) rapidly settle the child into bed; 5) leave the bedroom; 6) ignore child protestations unless in case of illness; 7) if child leaves bedroom after settling time they are firmly told to return and, if necessary, physically carried back with minimal affective contact. **Duration:** 2 weeks **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):** Single face-to-face session at the clinic or participant's home to explain the treatment. There was brief telephone contact on the following three days to offer encouragement and deal with any problems. There was additional telephone contact if necessary. Based on a review of case notes the author states that only a minority required more than 4 phone calls and in only one case was there more than 7. **Description of comparator:** No comparator		
The outcomes measures		
Outcome 1: Sleep problem severity **Details of measurement:** Parents rated their child's sleep severity on a visual analogue scale (VAS) ranging from zero (no problem) to 10 (severe problem). Measured at baseline, at the end of treatment and at follow-up. **Outcome 2:** Sleeping with parents **Details of measurement:** The number of children still sleeping with parents at follow-up **Outcome 3:** Frequency of night waking **Details of measurement:** Based on a nightly sleep diary completed by parents **Outcome 4:** Time to settle **Details of measurement:** Based on a nightly sleep diary completed by parent **Outcome 5:** Daytime behaviour problems **Details of measurement:** Children's daytime behaviour problems were assessed using the Behaviour Problem Index (Cunningham 1986) with a score range of 0 to 64. **Outcome 6:** Maternal Stress		

Details of measurement: Assessed using Rutter's Malaise Inventory (Rutter 1970) scoring from 0 (no problems) to 11.

Outcome 7: Maternal Sleep Scale

Details of measurement: Completed by mothers to rate their own sleep quality. Used an adapted version of Maternal Sleep Scale (De Diana 1976). Yes/No responses were required to 11 statements about sleep quality. Score range from 0 to 11 (better sleep quality).

Outcome 8: Helpfulness of the approach

Details of measurement: Parents rated the overall helpfulness of the treatment on a VAS ranging from zero (no help at all) to 10 (extremely helpful). Measured at end of treatment and at 4 month follow-up.

Outcome 9: Acceptability of approach

Details of measurement: Parents were asked to circle the phrase which best represented their view of the style of the treatment: 'too tough'; 'rather tough'; 'just right'; 'rather soft'; 'too soft'. Measured at 4 month follow-up

Length of follow-up: end of treatment; 4 months and 18 months after treatment

Summary of the results:

- Sleep problem severity - The mean severity reduced from 8 (SD 1.34; range 6 to 10) at baseline to 2.3 (SD 1.9, range 0 to 5) at end of treatment, 2.2 (SD 1.9, range 0 to 6) at 4 month follow-up and 2.9 (SD 2.2, range 0 to 6) at 18 month follow-up. ($p < 0.0001$; Friedman statistic 28.2; df3)
- Speed of change - Parents were asked how soon improvements in their child's sleep occurred. The mean number of nights within which change was observed was 3.6 (SD 1.9, range 1 to 7 nights)
- Sleeping with parents – At 4mth follow-up 10 of the 11 children who were regularly sleeping with their parents at baseline were no longer doing so.
- Frequency of night waking – Complete data not reported. There was a 59% reduction in the reported frequency of night waking in the cohort.
- Time to settle (based on data from 8 children) – There was a reduction in the mean time taken to settle from 58.6mins (SD 24.6) at baseline to 15.8mins (SD 7.8) at end of treatment and 17.5mins (SD 10.4) at 4 month follow-up.
- Daytime behaviour problems – There was a statistically significant improvement in daytime behaviour from baseline (mean 32.6, SE 3.5) to 4-month follow-up (mean 22.1 SE 3.2) ($p<0.01$)
- Maternal Stress (Malaise Inventory) – There was a statistically significant reduction in maternal stress over time: Baseline mean 8.7 (SE 1.1); end of treatment mean 4.7 (SE 1.0); 4-month follow-up mean 3.4 (SE 1.0) ($p<0.001$)
- Maternal Sleep Scale – maternal sleep quality improved over time: Baseline mean 4.1 (SE 0.6); end of treatment mean 7.1 (SE 0.6); 4-month follow-up mean 9.0 (SE 0.4) ($p<0.001$)

Any negative consequences: There were no reports of adverse effects

Views of parents:

- Acceptability of approach - 12 parents were of the view that the treatment approach was 'just right' for their children and 3 though it was 'rather tough'.
- Satisfaction with treatment - There was high overall satisfaction amongst parents with the treatment (at end of treatment the mean satisfaction score was 8.6 (SD 1.6)) and at 4 month follow-up it was 8.9 (SD1.9))

Authors' conclusion: The treatment approach was rapidly successful, well tolerated and acceptable.

Publication details		
Author: Colville[32]	**Year:** 1996	**Related publications:** Waiting on MSc thesis which contains full report
Stated aim: To establish whether standard behavioural techniques such as those commonly used with children under five years by psychologists and health visitors in primary health-care settings could help reduce the heavy burden on families.		
Study design: Before and after		
The participants		
Number: N=5	**Age:** 5yrs 1mth to 7yrs 8mths	**Sex:** 2 male, 3 female
Type of disability: Sanfilippo syndrome (4 sub-type A, 1 sub-type B)		
Sleep problem: Bedtime disturbance, night waking and disruption **How the sleeping problem was assessed:** Questionnaire based on Richman and Graham (1986)		
The intervention		
Setting: Home-based **Type of behavioural intervention:** Behavioural intervention **Description of intervention:** **Duration:** 6 weeks **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):** Home visit by clinical psychologist before and during the intervention period to negotiate the treatment plan. Weekly telephone contact throughout the treatment period. **Description of comparator:** No comparator		
The outcomes measures		
Outcome 1: Goal achievement **Details of measurement:** Whether or not the treatment goal had been achieved **Length of follow-up:** End of treatment and 4 months after intervention started		
Summary of the results: There were two treatment goals for four children and three for the fifth child. For three of the four both treatment goals were achieved at the end of treatment, for the fourth child neither were achieved and for the fifth child two of the three goals were achieved. Follow-up data were available for three children: for one child both goals were maintained, for one child neither was maintained and one was maintained for the final child. **Any negative consequences:** **Views of parents:**		
Authors' conclusion: The results of the interventions were encouraging.		
Comments Full data not reported in this paper. Waiting on full report.		

Publication details		
Author: Christodulu[27]	**Year:** 2004	**Related publications:**
Stated aim: To investigate the effectiveness of positive bedtime routines and sleep restriction in reducing bedtime disturbances and night awakenings in children with developmental disabilities		
Study design: Before and after		
The participants		
Number: N=4	**Age:** 2yrs 6mths; 2yrs 9mths; 3yrs 11mths; 5yrs 11mths	**Sex:** 2 male, 2 female
Type of disability: Developmental disabilities (CHARGE association; pervasive developmental disorder, sensory integration and hypotonia; immune deficiency; autism)		
Sleep problem: Bedtime disturbances and night wakening. All of the children had an irregular sleep schedule with variation from night to night in bedtime and wakening time. **How the sleeping problem was assessed:** The Albany Sleep Problems Questionnaire was used to assess type and severity of sleep disturbance; the Sleep Intervention Questionnaire (designed for the study) to assess the appropriateness of using sleep restriction; the Parental Sleep Satisfaction Questionnaire (PSSQ); and parents were also interviewed and completed daily sleep charts and bedtime behaviour logs.		
The intervention		
Setting: Home-based **Type of behavioural intervention:** Positive bedtime routine and sleep restriction (sleep restriction only for one child) **Description of intervention:** 1) *Positive bedtime routine* - this was introduced prior to the introduction of sleep restriction. Parents were asked to create a routine that they could follow based on the following guidelines: a) have a regular routine in the 30mins before bedtime; b) include activities such as washing, putting on sleepwear and reading; c) keep the order and timing of the activities about the same each evening; d) do not include activities that could cause conflict; e) avoid watching television; f) avoid extending the length of the routine. 2) *Sleep restriction* – The amount of time the child was in bed was restricted to 90% of the total time that the child slept (based on parent sleep diaries). The child's bedtime and/or the time the child was woken were adjusted for the new schedule. **Duration:** 1) The positive bedtime routine phase lasted from a few days to approximately 6 weeks. 2) The sleep restriction plus positive bedtime routine phase lasted approximately 14-18 weeks **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):** Details not provided **Description of comparator:** No comparator		
The outcomes measures		
Outcome 1: Total sleep time **Details of measurement:** Based on parental sleep diaries **Outcome 2:** Number and duration of bedtime disturbances **Details of measurement:** Based on parental sleep diaries **Outcome 3:** Night wakening **Details of measurement:** Based on parental sleep diaries		

Outcome 4: PSSQ

Details of measurement: Created for the study to assess parental satisfaction with their child's current sleep pattern. Score ranges from 6 (less satisfaction) to 36.

Length of follow-up: End of treatment and one month follow-up

Summary of the results:

- Total Sleep Time – This decreased for three of the 4 children by 30 to 90 minutes following the intervention. The sleep restriction phase was not implemented for one child due to illness and the total time sleeping did not change from baseline
- Bedtime disturbances – There was a reduction in the frequency and duration of bedtime disturbances for all 4 children.

  Child 1 – Decreased from a mean frequency at baseline of 4.22 disturbances (range 2 to 7) per week to 0.00 (range 0) at follow-up and a mean duration of disturbances of 245mins per week (range 75 to 420) to 0mins (range 0)

  Child 2 - Decreased from a mean frequency at baseline of 6.62 disturbances (range 2 to 7) per week to 0.50 (range 0 to 1) at follow-up and a mean duration of disturbances of 849mins per week (range 435 to 1,525) to 30mins (range 0 to 60)

  Child 3 (bedtime routine only)- Decreased from a mean frequency at baseline of 6.5 disturbances (range 4 to 7) per week to 2.5 (range 2 to 3) at follow-up and a mean duration of disturbances of 232mins per week (range 85 to 295) to 75mins (range 75)

  Child 4 - Decreased from a mean frequency at baseline of 3.10 disturbances (range 1 to 6) per week to 0.50 (range 0 to 1) at follow-up and a mean duration of disturbances of 88mins per week (range 15 to 420) to 23mins (range 0 to 45)
- Night Wakening - There was a reduction in the frequency and duration of night wakening for all 4 children.

  Child 1 – Decreased from a mean frequency at baseline of 3.44 awakenings per week (range 1 to 7) to 0.05 (range 0 to 1) at follow-up and a mean duration of awakenings of 291mins per week (range 50 to 545) to 10mins (range 0 to 20)

  Child 2 – Decreased from a mean frequency at baseline of 8.27 awakenings per week (range 4 to 12) to 4.00 (range 4) at follow-up and a mean duration of awakenings of 682mins per week (range 280 to 1,180) to 278mins (range 275 to 280)

  Child 3 (bedtime routine only) – Decreased from a mean frequency at baseline of 9.29 awakenings per week (range 8 to 12) to 1.50 (range 1-2) at follow-up and a mean duration of awakenings of 92mins per week (range 52 to 180) to 8mins (range 5 to 10)

  Child 4 – Decreased from a mean frequency at baseline of 1.70 awakenings per week (range 0 to 4) to 1.00 (range 0 to 2) at follow-up and a mean duration of awakenings of 258mins per week (range 0 to 562) to 120mins (range 0 to 240)
- PSSQ – Parental satisfaction with their child's sleep increased from baseline to follow-up

  Child 1 – Mean score increased from 14.67 (range 11-19) at baseline to 21.50 (range 18-25) at follow-up

  Child 2 - Mean score increased from 6.67 (range 6-7) at baseline to 24.00 (range 24) at follow-up

  Child 3 - Mean score increased from 11.71 (range 10-15) at baseline to 24.00 (range 24) at follow-up

  Child 4 - Mean score increased from 14.00 (range 12-16) at baseline to 28.00 (range 28) at follow-up

Any negative consequences: The authors state that the children did not experience any adverse consequences due to the decreased sleep time. **Views of parents:** The authors state that the parents found the intervention easy and practical to implement.
Authors' conclusion: The results support the use of sleep restriction, in conjunction with positive bedtime routines, for the treatment of sleep problems in children with developmental disabilities.
Comments: Although reduction, some children still had disturbance/wakening

Publication details		
Author: Didden[21]	**Year:** 2004	**Related publications:**
Stated aim: To assess the effectiveness of functional assessment and behavioural treatment of sleep problems in children with developmental disability.		
Study design: Before and after		
The participants		
Number: N=3	**Age:** 9.2, 10 and 12.4yrs	**Sex:** 3 males
Type of disability: Moderate developmental disability with Downs Syndrome; seizure disorder; mild developmental disability with ADHD (taking Ritalin)		
Sleep problem: One displayed disruptive behaviour at bedtime and would only sleep if one of his two carers lay in bed with him until morning; and two had night wakening **How the sleeping problem was assessed:** Functional assessment based on parental interview and nightly recordings made by parents over one week that recorded each night antecedent and consequent event and number of minutes of disruptive behaviours.		
The intervention		
Setting: Home-based **Type of behavioural intervention:** Extinction for two children; differential reinforcement of incompatible behaviours (DRI) using tokens plus response cost for one child **Description of intervention:** 1) *Extinction* - Parents were asked to follow a bedtime routine. Toys were removed from the bedroom to prevent play during the night. After putting child to be and saying goodnight they had to leave the room and were instructed not to re-enter the room until morning. When illness was suspected they could re-enter but attention was kept to a minimum. When the child slept through the night they were told that because they had been quiet during the night they had earned extra positive attention in the morning. 2) *DRI plus response cost* – The child was given 10 tokens at bedtime and one token was taken away each time he showed disruptive behaviours. Five tokens by morning earned a preferred activity (e.g. playing Gameboy). After three consecutive nights earning a preferred activity the number of tokens required was increased by one. Extinction was then added and tokens were removed without any comment. Because these procedures were not effective a punishment was added: if 5 tokens or more were lost his bedroom door was locked for the rest of the night. **Duration:** Approximately 40 nights and 80 nights for extinction and 80 nights for DRI **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):** There was daily phone contact with parents. The authors state that this was an important part of the treatment programme especially during initial treatment. **Description of comparator:** No comparator		
The outcomes measures		
Outcome 1: Number of minutes of night-time disruption **Details of measurement:** Defined as any disruption (e.g. out of bed, hitting, kicking objects) of at least one minute between sleep time and wake time. Recorded by parents on a standardised sheet nightly. **Length of follow-up:** End of treatment and 6mths after treatment		
Summary of the results: • Night-time disruption – Decreased in all three children.		

Child 1 – Decreased from mean 44.1mins (SD 12.9, range 24-65) at baseline, to 11.1 (SD 15.7, range 0-59) during treatment and 0.3 (SD 0.5, range 0-1) at follow-up

Child 2 - Decreased from mean 131.4mins (SD 139.2, range 0-405) at baseline to 62.9 (SD 60.5, range 0-319) during treatment and 0.12 (SD 9.2, range 0-20) at follow-up.

Child 3 - Decreased from a mean of 65.2mins (SD 59.8, range 0-165) at baseline, to 48.5 (SD 20.3, range 03-83) during response cost and DRI, 49.8 (SD 28.4, range 0-90) during response cost, DRI and extinction, 23.1 (SD 28.1, range 0-121) during response cost, DRI, extinction and punishment and 12.6 (SD 14.2, range 1-34) at follow-up.

Any negative consequences: None reported

Views of parents: The authors state that the parents found it difficult to implement the intervention initially but continued on the program and were 'highly contented' with the results.

Authors' conclusion: The results demonstrate the effectiveness of functional assessment and behavioural treatment of severe sleep problems in three children with developmental disability.

Publication details		
Author: Didden[22]	**Year:** 2002	**Related publications:**
Stated aim: To assess the effectiveness of extinction of parental attention (planned ignoring) on night-time disruptive behaviours.		
Study design: Before and after		
The participants		
Number: N=4	**Age:** 1yr, 11mths; 7yrs, 3mths; 6yrs, 5mths; 25yrs;	**Sex:** 3 males, 1 female
Type of disability: Two with severe learning disabilities, one moderate to severe learning disabilities and one with mild delays in several developmental areas.		
Sleep problem: One went to bed willingly but woke several times during the night and behaved disruptively by screaming and yelling; one had problems settling as well as disruptive behaviours during the night; one refused to go to bed most nights and slept in parents bed most nights; one had problems settling and frequently woke during the night and cried. **How the sleeping problem was assessed:** Functional assessment based on interview with parents and nightly completion by parents of a form recording antecedent and consequent events and number of minutes of night-time disruptive behaviours.		
The intervention		
Setting: Home-based **Type of behavioural intervention:** Extinction **Description of intervention:** Parents were instructed to discontinue their usual management techniques. They were asked to put the child to bed, say 'good-night' and after leaving the bedroom not to re-enter until morning. In the case of illness they could re-enter the room but were asked to keep interaction to a minimum. When the child slept throughout the night they explained to him/her that they had earned positive attention during the morning because they had been quiet during the night. **Duration:** Varied across participants – ranged from to extinction periods of 10 nights each to an extinction period of 120 nights (figures approximate from graph) **If delivered by parents, give description of training and support received**: Not explicitly stated, though the authors advise daily contact between the therapist and parents particularly during the first week of the intervention **Description of comparator:** No comparator		
The outcome measures		
Outcome: Night-time disruption (any disruption by the child for at least one minute - such as crying, screaming, getting out of bed – between time of settling to sleep and wake-up time) **Details of measurement:** Measured nightly by one parent using a standardised form. Measured at baseline, during treatment and follow-up. **Length of follow-up:** end of treatment and 6 months after treatment		
Summary of the results: • 7yr, 3mth old with severe learning disabilities – The mean number of minutes of disruption reduced from 45.4mins (SD 29.2) at baseline to 15.9mins (SD 31.9) during treatment and 3.8mins (SD 7.5) at follow-up. • 6yr, 5mth old with moderate to severe learning disabilities – The mean number of minutes of disruption reduced from 26.8mins (SD 20.9) at baseline, 32.4mins (SD 28.2) during		

treatment to 1.1mins (SD 2.1) at follow-up. • 1yr, 11mth old with mild developmental delays – The mean number of minutes of disruption were 1min (baseline 1); 28.7mins (SD 32.7) (extinction 1); 1min (baseline 2); 1.5mins (SD 3.2) (extinction 2); 0.4mins (SD 1.1) (follow-up) (there may be an error in these data as the pattern is very different to the other two children) **Any negative consequences:** There was a temporary increase in night-time disruptive behaviour during initial treatment sessions in one child. **Views of parents:** The authors state that parents found it difficult to implement the intervention during the initial treatment sessions. The parents had concerns about causing psychological trauma to their child and that the child might experience feelings of rejection and fear.
Authors' conclusion: Treatment resulted in a normalised sleep pattern in all cases and effects were maintained across time.

Publication details		
Author: Didden[23]	**Year:** 1998	**Related publications:**
Stated aim: To assess the effectiveness of several procedures on sleeping problems with six developmentally delayed disabled children at young age who live at home		
Study design: Before and after		
The participants		
Number: N=3 (The study included 6 children but 1 had night terrors and 1 had sleep problems related to seizures. Before and after data were available for 3 of the remaining 4)	**Age:** 2, 4 and 6 yrs	**Sex:** 3 male
Type of disability: Spinal muscle atrophy, ADHD (both near normal IQ), Prader-Willi syndrome		
Sleep problem: Problems settling, night waking and co-sleeping with parents **How the sleeping problem was assessed:** Functional assessment based on interview with parents and nightly completion (6 nights) of standardised sleep diary recording antecedent and consequent events and duration of night-time disruptive behaviours.		
The intervention		
Setting: Home-based **Type of behavioural intervention:** Extinction (non-graduated) **Description of intervention:** Parents were instructed to discontinue their usual management techniques. They were asked to put the child to bed, say 'good-night' and after leaving the bedroom not to re-enter until morning. In the case of illness they could re-enter the room but were asked to keep interaction to a minimum. When the child slept throughout the night they explained to him/her that they had earned positive attention during the morning because they had been quite during the night. **Duration:** Varied across participants – approximately 50 nights, 54 and 29 nights **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):** Not explicitly stated **Description of comparator:** No comparator		
The outcomes measures		
Outcome 1: Night-time disruption (any disruption by the child for at least one minute - such as crying, screaming, getting out of bed – between time of settling to sleep and wake-up time) **Details of measurement:** Measured nightly by one parent using a standardised form. Measured at baseline, during treatment and follow-up. **Length of follow-up:** End of treatment and 3 months after treatment for child 1 and 6 months after for child 2 and 3		
Summary of the results: • 2 year old with spinal muscle atrophy - The mean number of minutes of disruption reduced from 131mins at baseline to 0mins by the sixth night of treatment • 4 year old with Prader-Will syndrome - The mean number of minutes of disruption reduced from 90mins (range 45 to 180) at baseline to 22mins (range 5 to 180) during treatment to		

0mins at follow-up. • 6 year old with ADHD - The mean number of minutes of disruption reduced from 21mins (range 9 to 27) at baseline to 9mins (range 0 to 26) during treatment and 1.7mins (range 0 to 4) at follow-up. **Any negative consequences:** None reported **Views of parents:** None reported
Authors' conclusion: Behavioural procedures may be effective in decreasing sleeping disorders with young developmentally disabled children

Publication details		
Author: Durand[28]	**Year:** 2004	**Related publications:**
Stated aim: To investigate the effectiveness of sleep restriction in reducing bedtime disturbances and night wakening in two children with developmental disabilities		
Study design: Before and after		
The participants		
Number: N=2	**Age:** Both 4yrs	**Sex:** 2 females
Type of disability: One with autism and one with developmental delays		
Sleep problem: One child with night wakening and getting into bed with parents and frequent crying and not getting back to sleep. This child also had severe bedtime disturbances which, at baseline were controlled with melatonin. One child with severe bedtime disturbances and periodical night wakening. **How the sleeping problem was assessed:** The Albany Sleep Problems Questionnaire was used to assess type and severity of sleep disturbances and the Parental Sleep Satisfaction Questionnaire (PSSQ) (Christodulu, 2000) to assess parental satisfaction with the child's current sleep pattern. Parents were also interviewed and completed nightly sleep charts. Sleep restriction was used because extinction had previously been unsuccessful.		
The intervention		
Setting: Home-based **Type of behavioural intervention:** Sleep restriction and consistent bedtime routines and practices **Description of intervention:** 1) Sleep restriction – The amount of time the child was in bed was restricted to 90% of the total time that the child normally slept at baseline (based on parent sleep diaries). The child's bedtime and/or the time the child was woken were adjusted for the new schedule. 2) Parents were instructed to establish consistent bedtime routines and ways of responding to bedtime disturbances and wakening. These included not getting into bed with the child or allowing the child to get into the parental bed. If the child left their bed they had to return her to her own bed, tell her to go to sleep and leave the bedroom. **Duration:** Approximately 15 and 25 weeks **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):** Not reported **Description of comparator:** No comparator		
The outcomes measures		
Outcome 1: Total Sleep Time **Details of measurement:** Based on parental sleep diaries. **Outcome 2:** Number and duration of bedtime disturbances **Details of measurement:** Based on parental sleep diaries. **Outcome 3:** Number and duration of night wakening **Details of measurement:** Based on parental sleep diaries. **Outcome 3:** PSSQ **Details of measurement:** To assess parental satisfaction with their child's current sleep pattern. Score ranges from 6 (less satisfaction) to 36. **Length of follow-up:** End of treatment		
Summary of the results: • Total Sleep time – Decreased from 8.75hrs per night at baseline to 7hrs during the		

intervention for the first child and from 10.85hrs per night at baseline to 9.5 during the intervention for the second. The authors state that when the programme was successful the amount of sleep was faded back to an age appropriate level.

- Bedtime disturbances
  Child 1 - The melatonin used at baseline was effective in controlling bedtime disturbances. When the sleep restriction was introduced the melatonin was withdrawn without any return to bedtime disturbances.
  Child 2 - Decreased from a mean frequency of 7 disturbances (range 7) per week at baseline to 0.25 (range 0-1) following intervention. Mean duration decreased from 1.05hrs per week (range 0.79-1.35) at baseline to 0.01 hrs (range 0-0.04) following intervention.
- Night wakening – The frequency and duration reduced for both children
  Child 1 – Decreased from a mean frequency of 7.17 wakings per week (range 5-9) at baseline to 1.43 (range 0-4) per week following intervention. Duration decreased from a mean of 1.27hrs per week (range 0.18-2.2) at baseline to 0.18hrs per week (range 0-1.11) following intervention.
  Child 2 - Decreased from a mean frequency of 2.55 wakings per week (range 0-6) at baseline to 1.38 (range 0-3) per week following intervention. Duration decreased from a mean of 0.14hrs per week (range 0-0.37) at baseline to 0.07hrs per week (range 0-0.15) following intervention.
- PSSQ – Parental satisfaction with their child's sleep increased from baseline to follow-up.
  Child 1 – Mean score increased from 6 at baseline to 23 following treatment
  Child 2 – Increased from 8 at baseline to 30 following treatment

Any negative consequences: Child 1 experienced an increase in sleep walking by the third week of the intervention (mean 2.3 episodes per week). These decreased as the sleep time was extended. This child also experienced two episodes of sleep terrors during the intervention.
Views of parents: The authors state that the parents thought it was easy to implement sleep restriction on a regular basis.

Authors' conclusion: The results support the use of sleep restriction for the treatment of sleep disturbances in children with developmental disabilities.

Publication details		
Author: Durand[24]	**Year:** 1996	**Related publications:**
Stated aim: To evaluate the effectiveness of behavioural interventions, including graduated extinction in reducing night wakening and bedtime disturbance in children with autism and other developmental disabilities		
Study design: Before and after		
The participants		
Number: N=4	**Age:** 2, 7, 11 and 12 years old	**Sex:** 2 male, 2 female
Type of disability: Two with mild to moderate learning disabilities, one with pervasive developmental delays and one with autism and challenging behaviours.		
Sleep problem: Two had frequent night-time wakening and two had disruptive behaviour at bedtime **How the sleeping problem was assessed:** The Albany Sleep Problems Questionnaire was used to assess type and severity of sleep disturbance. Parents were also interviewed and completed nightly sleep charts by parents.		
The intervention		
Setting: Home-based **Type of behavioural intervention:** Graduated extinction (and establishment of consistent bedtime routine) **Description of intervention:** A consistent bedtime routine was established for each child; the timing and nature of the routine varied between children depending on their needs. When children were disruptive during the night only neutral reassurance ('It is still time to sleep, go back to sleep') was given and physical contact kept to a minimum. Parents were instructed not to get into their child's bed during the night or to allow the child into their bed. The graduated extinction schedule in response to night wakening or disruptive behaviour varied between children: 1) parent started with waiting 3 minutes before entering bedroom and this increased by 2 minutes each night to a maximum of 10 minutes; 2) parent started with a 5 minute delay which increased by 5 minutes each night; 3) parent started with 3 minute delay increasing by 2 minutes each night; 4) no incremental delay **Duration:** 8 to 16 weeks (for one child formal assessment was 2 weeks as she developed an illness) **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):** The authors state that there was regular telephone contact with parents during the baseline and treatment sessions. **Description of comparator:** No comparator		
The outcomes measures		
Outcome 1: Night wakening **Details of measurement:** Based on daily sleep charts completed by parents. Reported as the percentage of nights per week with waking or disturbance. **Outcome 2:** Bedtime disturbances **Details of measurement:** Based on behaviour logs completed daily by parents. Reported as the percentage of nights per week with bedtime disturbances.		

Length of follow-up: end of treatment and for one participant there was follow-up at 2 and 6 months and for one at 1 and 2 months post-treatment.

Summary of the results:

- Night wakening – there was a reduction in the % of nights with night wakening per week for the two children with this problem. In one child this decreased from a mean of 36.4% (range 14.3 to 57.1) at baseline to 11.4% (range 0 to 28.6) during treatment; in the second child the decrease was from a mean of 93.6% (range 71.4 to 100) at baseline to 64.3% (range 57.1 to 71.4) during treatment, 50% at 2 months follow-up and 26.8% (range 25 to 28.6) at 6 months follow-up. Other behaviours that were a target of the intervention also showed improvement: the first child had a more regular bedtime and the mother of the second child no longer stayed in bed with her following awakenings.
- Bedtime disturbances - there was a reduction in the % of nights with bedtime disturbance per week for the two children with this problem. In one child this reduced from a mean of 100% at baseline to 22.3% (range 0 to 66%) during treatment; in the second child the decrease was from a mean of 65.1% (range14 to 100) at baseline to 22.3% (range 0 to 100) during treatment, 14% at 1 month and 0% at 2 months follow-up. The mean length of time to fall asleep for this child reduced from 133.3 minutes (range 50.7 to 233.6) to 44.4 minutes (range 0 to 162.9).

Any negative consequences: None reported

Views of parents: The authors state that parents were at first hesitant to delay attending to their children but found the short delay easy to tolerate.

Authors' conclusion: The results of the study support the use of behavioural interventions for night wakening and disruptive bedtime behaviour in children with developmental disabilities.

Publication details		
Author: Hewitt[18]	**Year:** 1985	**Related publications:**
Stated aim: To describe the application and effectiveness of behavioural treatment of sleeplessness in a sample of 10 children with severe learning difficulties		
Study design: Before and after		
The participants		
Number: N=10	**Age:** Mean 6yrs 11mths (range 3yrs 2mths to 16yrs 6mths)	**Sex:** 8 male, 2 female
Type of disability: Severe learning difficulties (7 Downs Syndrome, 1 Cornelia de Lange syndrome, 1 tuberous sclerosis and one of non-specific origin)		
Sleep problem: 4 night-time wakening, 1 bedtime disturbances, 3 with both, 1 with repeated waking plus head-banging while awake and asleep and 1 child that had occasional episodes of staying awake all night. **How the sleeping problem was assessed:** There was a joint initial interview between families and a clinical psychologist and community nurse in the family home. Sleep patterns were recorded by parents for a one week baseline period using a 24-hour chart. **Other information:** The children were identified from 29 referred to a clinical psychology department for behavioural problems, whose parents thought sleeping problems was the main difficulty.		
The intervention		
Setting: Home-based **Type of behavioural intervention:** Positive bedtime routine and conditioning; the precise intervention was tailored to the individual needs and resources of each family **Description of intervention:** A tailored behavioural treatment programme was developed and negotiated with each family which was written up on the weekly chart. The following general framework was used: 1) positive bedtime routine that included set bedtime, introduction of a regular routine before bedtime that provided clear stimuli for the child that bedtime was approaching, avoidance of overstimulation in the hour before bed; 2) teaching a relaxation response after getting into bed through use of a bedtime story or soft music; 3) gradual distancing of parent from bedroom once relaxation response was established; 4) identification of factors that were maintaining disruptive behaviours and advice for more constructive parent responses. During wakeful episodes the stimulus to which the child had become conditioned to fall asleep was repeated. Parents were advised to interact with the child as little as possible and avoid prolonged routines and overstimulation during waking episodes. Parents were made aware of the importance of consistency and the possibility that progress may be slow. **Duration:** Mean 6.7 weeks (range 2-15 weeks). Parents were asked to stop recording sleep behaviour when the child settled easily at night and/or no longer woke at night or the parent's sleep was less disrupted. Recording could also stop if difficulties were only occasional and this was considered a satisfactory outcome. **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):** Following the assessment period which consisted of two visits to the family home by a clinical psychologist and community nurse, the nurse monitored the child's progress on a regular (usually		

weekly) basis. The psychologist also visited at three-weekly intervals and gradually withdrew visits as progress occurred. More complex cases received joint visits. There were monthly case review meetings.

Description of comparator: No comparator

The outcomes measures

Outcome 1: Brief summary of whether improvement occurred based on time to settle and frequency of night waking.

Details of measurement: Based on parental sleep recordings

Length of follow-up: End of treatment and approximately one year later

Summary of the results:

At baseline the average time taken to settle to sleep ranged from 34 minutes to 2.5hrs and the frequency of night waking from 6 to 18 episodes during the week. Following treatment eight of the 10 children showed a positive outcome: parents reported the children settling easily and/or no or only occasional night-time wakening. The mean length of time to a positive outcome was 6.7 weeks (range 2-15 weeks). One child did not receive behavioural treatment as it was established from the charts that there was a possible link with epilepsy. The child with repeated waking plus head-banging episodes did not improve. At one year follow-up 6 of the 8 maintained the improvement. Three had a slight relapse following a period of illness or disruption to the family routine. A regular sleeping pattern was re-established by parents with a minimum of professional involvement.

Any negative consequences: None reported

Views of parents: The authors state that some parents viewed sleeplessness as being directly attributable to their child's disability. It was important to 'sell' a behavioural approach prior to the intervention to these parents.

Authors' conclusion: The authors make a number of observations: they highlight that many programme modifications were necessary to ensure the individual interventions suited individual parenting styles and family resources; they state that it was not possible to identify the elements of the intervention that were most important and that in addition to the specific techniques factors such as directly involving parents, a written treatment programme, daily feedback for parents from recordings and weekly support visits may have been important.

Publication details		
Author: Montgomery[14]	**Year:** 2004	**Related publications:**
Stated aim: To investigate the efficacy of a media-based brief behavioural treatment of sleep problems in learning disabled children by comparing treatment delivered face-to-face to control and treatment delivered by booklet to control		
Study design: Randomised controlled trial		
The participants		
Number: N=66	**Age:** 2 to 8 years	**Sex:** 42 male, 24 female
Type of disability: Severe learning disability (32% autism, 12% Down's Syndrome, 8% global developmental delay, 6% epilepsy, 21% other, 27% no diagnosis)		
Sleep problem: Night waking and/or settling problems. For entry into the trial children had to have severe sleep disturbance of at least 3 months duration unrelated to a physical problem. Severe problem was defined as night waking 3 or more times per week for more than a few minutes and disturbing parents or going into their room and/or problems settling 3 or more times per week where the child takes more than an hour to settle and causes disturbance during this time. **How the sleeping problem was assessed:** A brief screening questionnaire was used (Two papers are referenced regarding reliability and validity): Composite Sleep Disturbance Score was calculated based on a parent completed sleep diary over a 2 week period. Each group received a 90 minute assessment visit when a sleep history was taken during a semi-structured interview. **Other information:** The parents of all 268 children attending a special school or receiving pre-school teacher counsellor services in Oxford, Berkshire and Buckinghamshire were contacted to participate in the trial. 184 responded of whom 102 met the entry criteria. 76 consented to participate of whom 10 then dropped out		
The intervention		
Setting: Home-based **Type of behavioural intervention:** 1) Behavioural intervention presented to parents face-to-face or 2) through a booklet. **Description of intervention:** 1) Face to face group – a single researcher spent approximately 90 minutes with parents in their own home explaining the techniques detailed below (a to g); 2) Booklet group - the second group were given a booklet detailing the same information. It was 14 pages long and also included cartoons and specifically addressed the needs of learning disabled children. Based on the Flesch Readability Test it was readable by someone educated up to 13 years old. Apart from the 90 minute assessment visit there was no contact with this group. The aim was to train parents in both groups in the same behavioural techniques. (Consistency was checked by comparing a selection of taped face-to-face sessions against the content of the booklet.) The topics covered were a) normal sleep: setting realistic expectations and explanation of the benefits of normal sleep, b) introduction to behavioural techniques in general (e.g. ignoring, consistency and reward systems), c) monitoring behaviour to devise the intervention, d) good sleep habits (e.g. clear routines, putting children to sleep while awake but drowsy), e) techniques for changing settling and waking problems (ignoring the child, checking briefly at increasingly linger intervals and with minimal contact, gradually decreasing physical contact) f) removing child from parents bed using settling techniques above, g) rewards for desirable behaviour. **Duration:** 6 weeks **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):** Not explicitly stated but there does not appear to have been any contact beyond that described above.		

Description of comparator: Waiting list control group.

The outcomes measures

Outcome 1: Composite Sleep Disturbance Score (CSDS)

Details of measurement: Derived from sleep diaries completed by parents over a 2 week period. Duration and frequency of settling and night waking problems were each scored from 0 to 2. This scale ranges from a minimum possible score of 0 (no sleep problems) to 8. In this study the minimum possible score for entry to the trial was 4. A random selection of CSDS were randomly cross-checked for consistency of scoring and agreement levels were greater than 95%

Outcome 2: Reduction in CSDS of at least 50% (responders)

Details of measurement: The cut-off was based on asking parents what was the minimum improvement that would make the intervention worthwhile: 83% said if the problem was reduced by half.

Outcome 3: Parental views about the booklet

Details of measurement: Rated from 0 to 4 on relevance, ease of understanding and usefulness. The minimum possible score was 0 (worst) and maximum 12 (best).

Length of follow-up: End of intervention and 6 month follow-up

Summary of the results:

- CSDS – there was a statistically significant difference in the main comparison across the three groups (face-to-face, booklet and control) post-treatment (H=34.174, df=2, p<0.001). Post-hoc comparisons indicated that each of the intervention groups showed a greater improvement on the CSDS compared to the control group. This improvement was maintained at 6 months follow-up.
  Baseline – face-to-face (n=20) mean 6.55 (SD 1.31); booklet (n=22) mean 6.18 (SD 1.46); control (n=24) mean 6.0 (SD 2.35)
  Post-treatment - face-to-face mean 2.4 (SD 1.93); booklet mean 2.55 (SD 2.76); control mean 5.75 (SD 1.54)
  6 month follow-up - face-to-face mean 1.89 (SD 2.02); booklet mean 2.08 (SD 2.89)
- 50% symptom reduction on CSDS – there were 15 'responders' versus 5 'non-responders' in the face to face group; 15 versus 7 in the booklet group and no responders for the control group. The waiting-list control group were randomised to treatment following the trial: there were 9 'responders' versus 3 'non-responders' in the face-to-face group and 8 versus 4 in the booklet group.
- Parental views on the booklet – 23 participants rated the booklet (this included the group in the main trial and those in the waiting list group that later received the booklet intervention). Parents found the booklet helpful and appropriate (mean score 10.17 (SD 1.87).

Any negative consequences: None reported

Views of parents: Not reported apart from views on the booklet

Other results: Sub-group analyses were conducted to investigate any variation in CSDS by sociodemographic characteristics (number of parents, number of siblings, social class). None were statistically significant.

Authors' conclusion: The study confirms the effectiveness of conventional behavioural treatment for sleep problems in children with learning disabilities and shows that brief delivery of this treatment using a booklet did not reduce its effect.

Comments: When applying the findings to outside the research setting need to bear in mind that the group given the booklet also spent 90 minutes visit with a member of the research team. Although this was for assessment purposes it may also have had a therapeutic effect. There is the possibility that using a booklet with no professional contact may not be as effective.
The authors note that although there was no statistically significant difference between groups at baseline the face-to-face group had slightly worse sleep problems which may have been clinically important.

Publication details		
Author: Piazza[29]	**Year:** 1997	**Related publications:**
Stated aim: To compare the efficacy of a faded bedtime with response cost treatment to bedtime scheduling in treating multiple sleep problems in learning disabled children		
Study design: RCT		
The participants		
Number: N=14	**Age:** Mean 7.8yrs (range 4 to 14)	**Sex:** Not stated
Type of disability: 6 had profound developmental disabilities, 4 severe, 1 moderate to severe, 2 moderate and 1 undetermined		
Sleep problem: Children were included in the study if they slept 90% or less of what would be expected for their age. The participants displayed a range of problems related to settling at bedtime and/or night-time waking. **How the sleeping problem was assessed:** Half-hourly observations over 24 hours **Other information:** The children had been admitted to the unit for displaying severe behaviour problems that posed a danger to self or others.		
The intervention		
Setting: In-patient unit specialising in the assessment and treatment of destructive behaviour problems. **Type of behavioural intervention:** Children were randomly assigned to one of two types of intervention (7 in each group): 1) Faded bedtime with response cost (FBRC); 2) Bedtime scheduling **Description of intervention:** 1) *Faded bedtime with response cost (FBRC)* – a bedtime at which sleep onset was highly likely with 15 minutes was set (half an hour later than the average time of sleep onset at baseline). A consistent bedtime routine was established. The child was not permitted to go to sleep before this time and was woken at a set time each morning. The response cost occurred if the child did not fall asleep within 15 minutes: they were removed from bed and kept awake for one hour (played with toys, watched TV etc). They were then returned to bed and this was repeated until the child was put to bed and fell asleep within 15 minutes. If the child fell asleep within 15 minutes of bedtime, bedtime was made half an hour earlier the next night. If they did not fall asleep it was made half an hour later. 2) *Bedtime scheduling* – the child was put to bed following a consistent bedtime routine, woken at the same time each morning and not allowed to sleep at other times unless a nap was age appropriate. If so there was a set nap time. **Duration:** Until the child was discharged from hospital which was on average 8 weeks. **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):** Not delivered by parents **Description of comparator:** See above		
The outcomes measures		
Outcome 1: Hours of disturbed sleep **Details of measurement:** Duration of inappropriate sleep (sleep outside appropriate sleep hours) plus the duration of time the child was awake when they should be asleep. The reliability of the observations was assessed by having two observers on 86% of the days. Inter-observer agreement was 98.2%. **Length of follow-up:** Varied depending on child's length of stay. The last 10 days of treatment were used.		

Summary of the results: There was a greater reduction in hours of disturbed sleep with FBRC than bedtime scheduling (F 6.66, df=1, p<0.026). At baseline the mean hours of disturbed sleep were 1.44hrs in the FBRC group and 1.37 in the bedtime scheduling group. Post-treatment they were 0.53hrs with FBRC and 1.10hrs with bedtime scheduling. **Any negative consequences:** None reported **Views of parents:** None reported
Authors' conclusion: Faded bedtime with response cost was superior to the bedtime scheduling procedure in reducing the number of hours of disturbed sleep.
Comments : In-patient setting – may not be generalisable to the home-setting

Publication details		
Author: Piazza[30]	**Year:** 1991	**Related publications:**
Stated aim: To determine whether the sleep problems of girls with Rett syndrome was amenable to a faded bedtime procedure		
Study design: Before and after		
The participants		
Number: N=3	**Age:** Two aged 8rs and one of 4yrs	**Sex:** 3 female
Type of disability: Rett syndrome		
Sleep problem: One with delayed sleep onset with disruptive behaviour and excessive daytime sleep; one with night waking and self-injurious behaviour; and one with night waking, crying and screaming and getting into parental bed. **How the sleeping problem was assessed:** Half-hour observations over 24hr period		
The intervention		
Setting: In-patient for 2 and home setting for one child (Child 3) **Type of behavioural intervention:** Faded bedtime with response cost **Description of intervention:** A bedtime was set at which sleep onset was highly likely within 15 minutes (half an hour later than the average time of sleep onset at baseline). A consistent bedtime routine was established. The child was not permitted to go to sleep before this time and was woken at a set time each morning. The response cost occurred if the child did not fall asleep within 15 minutes: they were removed from bed and kept awake for one hour (played with toys, watched TV etc). They were then returned to bed and this was repeated until the child was put to bed and fell asleep within 15 minutes. If the child fell asleep within 15 minutes of bedtime, bedtime was made half an hour earlier the next night. If they did not fall asleep it was made half an hour later. **Duration:** Not stated, presumably until discharge **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):** With the exception of one child, the intervention was not delivered by parents. The training and support received by the parents of this child was unclear. **Description of comparator:** No comparator		
The outcomes measures		
Outcome 1: % of appropriate sleep **Details of measurement:** Number of hours of sleep during the defined sleep period divided by total number of hours in the defined sleep period. Based on half hourly observations over 24hrs. **Outcome 2:** % inappropriate sleep **Details of measurement:** Number of hours sleep during the defined wake time divided by the total number of hours in the defined wake time. Based on half hourly observations over 24hrs. **Outcome 3:** Frequency and duration of night waking **Details of measurement:** Night waking defined as wake periods during sleep time preceded and followed by at least a 15 minute sleep episode. Based on half hourly observations over 24hrs. **Outcome 4:** Delay to sleep onset **Details of measurement:** The number of hours beyond the scheduled sleep time in which sleep occurred. Based on half hourly observations over 24hrs. The reliability of the observations was assessed for one child. Overall agreement was high. **Length of follow-up:** Not stated, until discharge		

Summary of the results:

- % appropriate sleep – Child 1 showed a marginal increase from an average of 87% at baseline to 90% following treatment; Child 2 increased from 69% at baseline to 75% following treatment; Child 3 increased from 81% at baseline to 92% following treatment.
- % inappropriate sleep – Child 1 decrease from 12% to 2%; Child 2 this was not a problem at baseline; Child 3 reduced from 15% to 7.2%.
- Frequency and duration of night waking – Child 1 not a problem at baseline; Child 2 frequency decreased from 1hr at baseline to 0.6hrs following treatment; Child 3 frequency decreased from 0.9 per night at baseline to 0.6 and duration from average of 1.8hrs per night at baseline to 0.5hrs.
- Delay to sleep onset – For child 1 who had this problem this decreased from 1.25 hrs at baseline to 0.5hrs post treatment.

Any negative consequences: None stated

Views of parents: Not reported

Authors' conclusion: The treatment used in the current investigation appeared to affect the various sleep related difficulties experienced by girls with Rett Syndrome. However, the small sample size and the variability in improvement across the children limit the generalisability of the findings.

Comments: In-patient setting for two children – may not be generalisable to the home setting. Some of the improvements may not be clinically meaningful.

Publication details		
Author: Piazza[31]	**Year:** 1991	**Related publications:**
Stated aim: To investigate the efficacy of a faded bedtime procedure for the treatment of paediatric insomnia		
Study design: Before and after		
The participants		
Number: N=4	**Age:** 3, 4, 13 and 19yrs	**Sex:** 2 male, 2 female
Type of disability: Profound learning disability		
Sleep problem: Met DSM III-R criteria for insomnia. Displayed a range of problems including problems settling, night waking, early waking and disruptive behaviours **How the sleeping problem was assessed:** Half hour observations over 24hr period. **Other information:** the children had been referred for the assessment and treatment of self-injury		
The intervention		
Setting: In-patient unit specialising in the assessment and treatment of severe behaviour disorders. One child was treated as an out-patient. **Type of behavioural intervention:** Faded bedtime with response cost (FBRC) **Description of intervention:** A bedtime was set at which sleep onset was highly likely within 15 minutes (half an hour later than the average time of sleep onset at baseline). A consistent bedtime routine was established. The child was not permitted to go to sleep before this time and was woken at a set time each morning. The response cost occurred if the child did not fall asleep within 15 minutes: they were removed from bed and kept awake for one hour (played with toys, watched TV etc). They were then returned to bed and this was repeated until the child was put to bed and fell asleep within 15 minutes. If the child fell asleep within 15 minutes of bedtime, bedtime was made half an hour earlier the next night. If they did not fall asleep it was made half an hour later. **Duration:** Not stated, presumably until discharge **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):** With the exception of one child, the intervention was not delivered by parents. The training and support received by the parents of this child was unclear. **Description of comparator:** No comparator		
The outcomes measures		
Outcome 1: % of intervals appropriate sleep **Details of measurement:** Number of sleep intervals occurring during the defined sleep period divided by the number of intervals of the defined sleep period. Based on half-hourly observations over 24 hours. **Outcome 2: %** of intervals of inappropriate sleep **Details of measurement:** Number of sleep intervals during the defined wake time divided by the total number of intervals of defined wake time. Based on half-hourly observations over 24 hours. **Outcome 3:** Frequency of night waking **Details of measurement:** Number of awake periods during defined sleep times that were preceded and followed by a sleep episode of at least 15 minutes. Based on half-hourly observations over 24 hours. The reliability of the observations was assessed by assessing the agreement between two observers for a proportion of the observations. Overall, agreement was high.		

Length of follow-up: End of treatment, for one child there was a 1 month follow-up post-discharge, for one child a one year follow-up and for 2 children no follow-up.

Summary of the results:

- Intervals of appropriate sleep – There were improvements for all participants, though in some instances these were very small: Child 1 increased from an average of 78% at baseline to 87% following treatment; Child 2 increased from 75.8% at baseline to 89.2% following treatment and 90% at one year (for this child the baseline and post-treatment assessment were conducted at home and the one year follow-up as an in-patient); Child 3 increased from 57% to 72%; Child 4 increased from 74% to 77% and 86% at one month follow-up.
- Intervals of inappropriate sleep – Child 1 these were zero at baseline and following treatment; Child 2 decreased from an average of 11.3% at baseline to 2.1% post-treatment and 0.36% at one year; Child 3 decreased from 9% to 0%; Child 4 decreased from 0.9% to 0%.
- Frequency of night waking – 3 children showed decreased night waking though some changes may not have been clinically significant. Child 1 decreased from an average of 0.3 wakings per night at baseline to 0 post-treatment; Child 2 decreased from 1.09 to 0.64 and 0.09 at one year; Child 3 from 0.3 to 0.2; Child 4 data not given.
- The frequency of climbing in and out of bed decreased for the child with this problem from a 100% of nights at baseline to 16% of nights at follow-up (mean 30, range 15 to 51 at baseline to mean 1.1, range 0 to 20). The frequency of being brought into parents bed decreased for the child with this problem (mean 84.3 to 45.4). At one year the frequency was less than once every 2 months.

Any negative consequences: None reported

Views of parents: The authors state that anecdotally, the parents reported a high degree of satisfaction with the outcome.

Authors' conclusion: Each patient benefited from the intervention

Comments: In-patient setting – may not be generalisable to the home setting. Some of the improvements may not be clinically meaningful

Publication details		
Author: Quine [19]	**Year:** 1991	**Related publications:** Quine[35]Quine[36]Quine [37]
Stated aim: To conduct an intervention trial with 25 families to assess whether training health professionals to teach behavioural techniques to parents of children with learning disabilities is effective in reducing children's sleep disturbance		
Study design: Before and after (for some of the measures the results were compared to an age-matched random sample of children with sleep problems from another district who had not sought or been offered treatment)		
The participants		
Number: N=25	**Age:** 1yr and 9mths to 21 years old	**Sex:** 17 male, 8 female
Type of disability: global developmental delay, cerebral palsy, Down's Syndrome, Steinert's disease, moderate and severe learning difficulties, microcephaly and developmental delay, autism, congenital rubella syndrome, Cri du Chat syndrome, right hemiplegia		
Sleep problem: Children were eligible for the study if they had night settling problems or night waking or limited sleep 3 or more times per week. **How the sleeping problem was assessed:** Interview with parents and two week sleep diary completed by the parents. **Other information:** The parents of all children attending Medway schools, social education centres and child assessment and care centres that ran playgroups for children with learning difficulties were approached. 40 families expressed an initial interest and 25 completed the programme. 1 dropped out during the programme and 14 dropped out before the intervention began (reasons provided).		
The intervention		
Setting: Home-based **Type of behavioural intervention:** Positive bedtime routine and conditioning; the precise intervention was tailored to the individual needs and resources of each family (based on Hewitt (1985) **Description of intervention:** A tailored behavioural treatment programme was developed and negotiated with each family which was written up on the weekly chart. The following general framework was used: 1) positive bedtime routine that included set bedtime, introduction of a regular routine before bedtime that provided clear stimuli for the child that bedtime was approaching, avoidance of overstimulation in the hour before bed; 2) teaching a relaxation response after getting into bed through use of a bedtime story or soft music; 3) gradual distancing of parent from bedroom once relaxation response was established; 4) identification of factors that were maintaining disruptive behaviours and advice for more constructive parent responses. During wakeful episodes the stimulus to which the child had become conditioned to fall asleep was repeated. Parents were advised to interact with the child as little as possible and avoid prolonged routines and overstimulation during waking episodes. Parents were made aware of the importance of consistency and the possibility that progress may be slow. **Duration:** Range 5 to 30 weeks. Parents were asked to stop recording sleep behaviour when the child settled easily at night and/or no longer woke at night or the parent's sleep was less disrupted. Recording could also stop if difficulties were only occasional and this was considered a satisfactory outcome. **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):** Following the assessment period progress was monitored by the health-visitor on a weekly basis. Frequency of home visits was agreed between the health-visitor and parent. Advice on maintaining improvement was given when a satisfactory outcome was reached and there was a follow-up		

appointment after 3 months.
The project was staffed by 12 health visitors who were each responsible for two families. All 12 attended a 3-day course on behavioural approaches to sleep disturbance delivered by an educational psychologist, a social psychologist, a clinical psychologist and a lecturer in social work experienced in role playing techniques.
Description of comparator: No comparator for sleep measures.

The outcomes measures

Outcome 1: Settling problems
Details of measurement: Number of minutes to settle. Based on sleep diary
Outcome 2: Night waking
Details of measurement: Number of times child woke each night and the number of minutes the child was awake. Based on sleep diary.
Outcome 3: Maternal satisfaction with settling and wake patterns
Details of measurement: Rated satisfaction on a 7-point scale (1 'not satisfied' to 7 'satisfied')
Outcome 4: Behaviour Problem Index
Details of measurement: Twenty items related to behaviour are rated to 0 (no or trivial difficulties) to 2 (marked difficulties) by the interviewer based on descriptions of behaviour from parents. Only items related to daytime behaviour were used.
Outcome 5: Maternal Responsiveness
Details of measurement: Checklist of 10 items to examine parental responses to sleep problems. Each item rated from 0 (never) to 4 (always). Internal reliability reported as high.
Outcome 6: Maternal Stress and Morale (Malaise Inventory)
Details of measurement: 24 item binary choice questionnaire adapted from Cornell Medical Index (Rutter *et al.* 1970). Scores of 5 or 6 were considered outside the normal range and a score of 7 or more as critical. Information provided on test-retest reliability and internal reliability.
Outcome 7: Irritability and smacking
Details of measurement: Appears to be frequency but unclear whether per day or per week.
Outcome 8: Judson Self-rating Scale
Details of measurement: Measures acceptance and adjustment of mother to child Judson and Burden 1980). 22 items are rated using a 7-point scale. Information provided on internal reliability.
Outcome 9: Problems Faced by Mothers of Children with Sleep Problems (Problem Inventory)
Details of measurement: Ten items scored from 0 (never a problem) to 4 (always a problem
Outcome 10: Mother's Perceptions of Self, Child and Husband
Details of measurement: 20, 14 and 16 items respectively rated on a 7-point scale
Length of follow-up: End of treatment (range 5 to 30 weeks) and 3 months from completion of treatment

Summary of the results:

- Settling problems (15 children) – the time taken to settle decreased from a mean of 111mins (range 45-180) at baseline to a mean of 20.4mins (range 5-60) after the intervention.
- Night waking (15 children) – The frequency of night waking decreased from a mean of 3.1 times per night (range 2.2-4) at baseline to a mean of 0.3 (range 0-1.3). The duration decreased from a mean of 70.2mins per night (range 30-120) to a mean of 3.2mins (range 0-15). Eight children did not sleep in their own bed between 4 and 7 times per week at baseline. Post-treatment this had stopped for seven children and occurred once a week for the eighth child.
- Maternal Satisfaction with Settling and Waking Problems – Satisfaction improved with settling from a mean of 2.2 (SD 1.7) at baseline to 6.3 (SD 1.1) after the intervention ($p<0.001$). Satisfaction improved with waking from a mean of 2.7 (SD 1.9) to 6.2 (SD 1.4) ($p<0.001$). There was no statistically significant change in the satisfaction of mothers in the

comparison group over the same time period.

- Behaviour Problem Index – Daytime behaviour improved from baseline (mean 13, SD 4.6) to post-treatment (mean 9.7, SD 4.3) (authors state this is statistically significant). There was no statistically significant change in the comparison over the same time period.
- Maternal Responsiveness – There was a decrease in the maternal responsiveness score from baseline to end of treatment indicating that mothers were more able to ignore inappropriate behaviour and reinforce appropriate behaviours (mean 22.4, SD 6.3) at baseline; mean 18.6 (SD 5.2) at end of treatment, $p<0.001$). There was no statistically significant change in the comparison group over the same time period.
- Maternal stress and morale – stress improved from baseline (mean 6.4, SD 4.1) to post-intervention (mean 3.8, SD 2.8) ($p<0.001$) and morale increased (mean 6.7, SD 2.2 to mean 7.6, SD 1.3) ($p<0.01$). There was no statistically significant change in the comparison group over the same time period.
- Irritability and smacking – There was a statistically significant improvement from baseline to post-treatment in feelings of irritability towards their child, frequency of smacking and fear of losing control and punishing their child too severely.
- Judson Self-rating Scale – Maternal acceptance of and adjustment to their child improved from baseline (mean 104.3, SD 16.2) to post-intervention (mean 128.4, SD 14.4) ($p<0.001$). There was no statistically significant change in the comparison over the same time period, though the baseline scores of the comparison group showed a more positive attitude to begin with.
- Problem Inventory – There was an improvement in the extent of the problems experienced by families from baseline (mean 20.3, SD 7.2) to post-treatment (mean 14, SD 6.9) ($p<0.001$). There was no statistically significant change in the comparison group over the same time period.
- Mothers Perceptions of Self, child and Husband – Positive feelings towards self, child and husband improved from baseline (mean 97.4, SD 14.2; mean 65.4, SD 8.8; mean 84.3, SD 10.2 respectively) to post treatment (mean 113.1, SD 16.7; mean 72.6, SD 9.9; mean 100.8, SD 14.7 respectively) ($p<0.001$)
- 3 months follow-up (based on 20 families) – 11/12 children with settling problems maintained the progress made and some improved further; 10/12 maintained their progress with night waking. Overall 17/20 had maintained progress or improved

Any negative consequences: None reported

Views of parents: Several parents provided positive comments on the intervention. Some mentioned that it was difficult to do at the beginning in terms of having to be consistent, believing that it could work or leaving their child to cry. Some commented on the usefulness of recording information in the sleep diaries and some commented on the importance of support from the health visitors.

Authors' conclusion: The study produced a remarkably clear cut set of results. The results showed that it is possible to radically improve children's sleep behaviour and that the improvements result in a number of positive changes in relationships within the family.

Comments

The authors highlight the risk of selection bias. They compared their cohort to an age-matched random sample of children with sleep problems in another health district, who had not been offered or sought treatment. The study cohort had a greater proportion of boys, were more likely to have had their problem since birth, were more difficult to manage and there was greater marital unhappiness and maternal irritability.

Care needs to be taken interpreting the comparisons with the comparison group. The statistical tests looked at change within each group rather than between group comparisons.

Publication details		
Author: Stores[15]	**Year:** 2004	**Related publications:**
Stated aim: To assess the effectiveness of a simple behavioural approach, based on instruction delivered to groups of mothers of young children with Down's Syndrome, in preventing or minimising sleep problems.		
Study design: RCT		
The participants		
Number: N=46	**Age:** Mean 2yrs 8mths (range 7mths to 4yrs 9mths)	**Sex:** 22 Male, 24 female
Type of disability: All had Down's Syndrome (details of severity of learning disability not available)		
Sleep problem: 65% (n=30) had at least one behavioural sleep problem: 14 bedtime settling problems, 26 night waking, 14 early morning waking and 7 sleeping in parental bed. Six children also had a sleep related breathing problem. 35% (n=16) did not have a sleep problem. **How the sleeping problem was assessed:** The Composite Sleep Problems Score and the Sleep-Related Breathing Problem Score were completed. **Other information:** Families with children aged 6mths to 5yrs were recruited from Oxfordshire Down Syndrome Service, the Hampshire Branches of Down's Syndrome Association, Downs Heart Group, health visitors, community paediatricians and child development centres. 77 eligible children were identified of whom 46 agreed to participate.		
The intervention		
Setting: Home-based. Mothers received instruction at a group session at the Oxford Down's Syndrome Resource Centre or the Down's Syndrome Educational Trust in Portsmouth. **Type of behavioural intervention:** One session of instruction and provided with booklet **Description of intervention:** There were separate sessions for mothers of children under 2.5 years and for those 2.5 to 5 yrs old. Small groups of about 5 mothers were brought together for the single instruction session. This lasted about 90 minutes including a discussion period of 30 minutes. The session consisted of information and advice about children's sleep and explaining behavioural techniques for encouraging good sleep habits such as establishing a positive bedtime routine, rewarding good behaviour, ignoring unwanted behaviour, gradual change. Case studies were used to illustrate the techniques. An illustrated booklet was provided (Encouraging Good Sleep Habits in Young Children with Down Syndrome). Both the instruction session and booklet had been piloted. **Duration:** One month **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):** No additional support was provided beyond the instruction session and booklet. **Description of comparator:** Waiting list control		
The outcomes measures		
Outcome 1: Composite Sleep Problem Score **Details of measurement:** Measures the frequency and duration of settling problems, night waking, early waking and sleeping in parental bed. The possible score range is from 0 (no problems) to 14. **Outcome 2:** Sleep-Related Breathing Problem Score (SRBPS) **Details of measurement:** Measure frequency of symptoms associated with sleep-related breathing problems. **Outcome 3:** Actometry (This is not reported for the intervention versus comparison group)		

Details of measurement: Wrist-watch device that measures basic sleep-wake patterns.
Outcome 4: Educational impact
Details of measurement: Knowledge of the Sleep of Young Children Questionnaire and Knowledge of Behavioural Principles as Applied to Children Questionnaire.
Outcome 5: Mother's evaluation on the instruction session and booklet
Details of measurement: Constructed for study.
Length of follow-up: One month and 6 months

Summary of the results:

- CSPS – Based on a 3x2 ANOVA there was no statistically significant main effect or interaction for time or group. Baseline: Intervention mean 2.83 (SD 3.41); Control 3.38 (SD 3.38). 1 month: Intervention 2.67 (SD 2.93); Control 3.5 (SD 4.02). 6 month: Intervention 2.08 (SD 2.35); Control: 4.38 (SD 3.86). There was a statistically significant difference between groups at 6mths based on a post-hoc test.
- SRBPS - Based on a 3x2 ANOVA was no statistically significant main effect or interaction for time or group (data provided in paper)
- Educational impact – At 1-month follow-up mothers in the intervention group scored significantly higher on both knowledge questionnaire that the control group.
- Mothers' evaluation of intervention (based on 18 responses) – The presentation was rated as very useful (16%), quite useful (61%) and not very useful (17%). The booklet was rated as very useful (17%), quite useful (50%) and not very useful (22%). All but 2 mothers who gave the lowest rating said it was because their child did not currently have a sleep problem; 2 had tried the advice without success. 94% said that the presentation and the booklet were easy to understand.

Any negative consequences: None reported
Views of parents: See above

Authors' conclusion: Group instruction offers some benefit regarding behavioural sleep problems but not for sleep-related breathing problems to which more attention should be given in children with Down Syndrome.

Comments: Participants with and without a problem were in one group for analysis – this reduces the likelihood of a reduction in sleep problems in the group as a hole post-intervention. The length of follow-up may have been insufficient to assess the effectiveness of the intervention as a prevention measure.

Publication details		
Author: Thackeray[25]	**Year:** 2002	**Related publications:**
Stated aim: To demonstrate the effectiveness of standard extinction for treating sleeping problems in children with an intellectual disability, to obtain data on the social validity of the intervention and to assess whether there are any benefits for daytime behaviour in the school setting.		
Study design: Before and after		
The participants		
Number: N=3	**Age:** 5yrs, 5yrs 6mths and 10 yrs	**Sex:** 3 male
Type of disability: one severe, one moderate and one mild intellectual disability		
Sleep problem: Child 1 would not fall asleep and had tantrums unless father present and if he woke during the night disturbed the household until his father helped him re-settle; Child 2 would not fall asleep unless mother present, woke three times per night and sometimes early morning waking; Child 3 needed his mother present to fall asleep and got into bed with parents or sister during the night. **How the sleeping problem was assessed:** Parents completed screening questionnaire and the Behavioural Evaluation of Disorders of Sleep (BEDS) questionnaire **Other information:** 156 families were invited to participate through recruitment at a Special Developmental School and a Special School in northern Melbourne, Australia. Children with an intellectual disability according to international criteria, difficulties in settling, night waking or co-sleeping, not on current sleep medication and no epilepsy were eligible. Four families expressed an interest and were invited to participate. One withdrew after the first intervention session as they were not ready to make changes to their child's sleeping arrangements		
The intervention		
Setting: Home-based Parent training took place at a university psychology clinic. **Type of behavioural intervention:** Standard extinction with positive bedtime routine, reinforcement, effective instructions and partner support. **Description of intervention:** Parents received an intensive two session training programme based on 5 Step Sleep Programme (McDonald and Patzold). The first two hour session covered behavioural reinforcers, instruction giving and bedtime routine. Parents planned an appropriate routine and treatment goals were established. Parents were asked to implement what they had learned following the session. Parent support strategies and standard extinction were introduced at the second session. Standard extinction involved explaining the rules to the child and after putting the child to bed leaving the room and ignoring all crying or calling out. If the child came out of their room the parents were instructed to take the child immediately back to bed with minimum contact with child. If the child complied the child received positive reinforcement in the morning. Parents were advised of the possibility of an extinction burst. Modelling and role-playing was used during the sessions and written information and parent checklists also provided. **Duration:** 7 weeks **If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):** In addition to the two training sessions parents received support by telephone from the therapist on at least three mornings after extinction was implemented as well as weekly phone calls during the		

rest of the programme. Including the pre-treatment and review sessions the therapist had six hours face-to-face contact with each family at the clinic.

Description of comparator: No comparator

The outcomes measures

Outcome 1: Goal Achievement Scale

Details of measurement: At the beginning of the programme parents identified two to four goals they wished to achieve in relation to their child's sleeping problem. They identified what they would consider total (100%) success for each goal. The level of success was assessed based on parent completed sleep diaries.

Outcome 2: Actigraph

Details of measurement: An Actiwatch was worn over five consecutive nights in each assessment period. One minute sample periods were used.

Outcome 3: Programme Evaluation Questionnaire

Details of measurement: Assessed parent satisfaction with outcomes, acceptability of the methods used, ease of understanding, ease of implementing the behavioural strategies and satisfaction with the therapist. They were also asked what they like most and least about the programme and what they would change.

Outcome 4: BEDS

Details of measurement: Parent completed questionnaire with 5 subscales

Outcome 5: Daytime behaviour

Details of measurement: 1) An observational checklist completed by trained observers for on-task behaviour and activity type and frequency counts of 4 target problem behaviours identified for each child; 2) teachers completed Developmental Behaviour Checklist – Teacher version; 3) a teacher-completed diary of child behaviour at lunchtime and after school; 4) a parent-completed diary of child behaviour before and after school.

Length of follow-up: End of treatment and 3 month follow-up

Summary of the results:

- Goal Achievement Scale – For three children, the goal of falling asleep independently every night was met with 100% success post-intervention and at 3-month follow-up (from 0 nights at baseline to 7 nights); for two children a goal was to fall asleep in own bed every night and this was met with 100% success at post-intervention and follow-up (from 4.3 and 6.3 nights at baseline to 7); for two children a goal was no co-sleeping on any night during the week and this was met with 100% success (from 1.5 nights and 7 nights at baseline to 0). For one child night waking showed some improvement post-intervention and 100% success was achieved at follow-up (from 2.2 nights at baseline to 0 at follow-up) and for one child there was no improvement (3 nights at baseline, 2.9 post-intervention and 3.9 at follow-up). For the later child there was a suggestion of sleep apnoea.
- Actigraph – Two children refused to wear it at follow-up. At end of treatment the duration of nighttime sleep increased from baseline for the three children by 53, 60 and 77 minutes
- BEDS – at baseline the 3 children had clinical or above average sleep problems which improved to normal levels for two children by follow-up and for one child did not change.
- Daytime behaviour – Based on parent and teacher ratings there were some small positive changes in behaviours for two children and a slight deterioration for the third. Based on the observational data each child showed improvement on a single behaviour but no others. Based on the DBC-T all three children showed a reduction in the total score but this was described as a convincing reduction for one child only.

Any negative consequences: Two children experienced an extinction burst. **Views of parents:** Program Evaluation Questionnaire – The three parents were very satisfied with the outcomes of the intervention and the techniques used, they thought the programme was very appropriate for their child and would strongly recommend it to a friend. They particularly liked the support received. Things they did not like were the Actiwatch and Ignoring their child when calling.
Authors' conclusion: The study demonstrated the effectiveness of standard extinction for treating settling, co-sleeping and night waking problems in children with intellectual disabilities and has high social validity. Support for behaviour change as a result of improved sleep was equivocal.

Publication details		
Author: Weiskop[26]	**Year:** 2005	**Related publications:**
Stated aim: To evaluate the effectiveness of extinction for treating parent-referred sleep onset and maintenance difficulties in young children with an autism spectrum disorder or fragile X syndrome.		
Study design: Before and after		
The participants		
Number: N=13	**Age:** Mean 5yrs 1mth (range 1yr 1mth to 9yrs 1mth)	**Sex:** 10 males, 3 females
Type of disability: 5 autism, 1 Asperger syndrome, 7 fragile X syndrome (FXS)		
Sleep problem: bedtime disturbances, sleeping in parental bed, night waking and disruptive behaviour **How the sleeping problem was assessed:** Interview with parents and functional assessment using parent completed sleep diary from at least a 2-week period. **Other information:** With the exception of one child all lived in two parent families and apart from four fathers all parents participated in the programme. Parents were recruited through an advertisement in a disability newsletter or by referral from their medical practitioner. Criteria for inclusion were that the parents perceived their child had a sleeping problem, the child was diagnosed with an autism spectrum disorder or FXS and did not have epilepsy. Children with autism had to be between 2yrs 6mths and 7yrs and not taking medication for sleep problems or daytime behaviours. The age and medication requirements were not applied to children with FXS due to difficulties in recruitment. The results are based on 10 children. One family withdrew due to child illness, one withdrew as the parent had family issues to attend to and one was not included because although he completed the intervention there were several interruptions to the intervention due to illness.		
The intervention		
Setting: Home-based. Conducted in metropolitan Melbourne, Australia **Type of behavioural intervention:** Positive bedtime routine, reinforcement, effective instructions, partner support and extinction **Description of intervention:** There were three weekly training sessions for parents. These covered the topics of goal setting (what they wanted to achieve with their own child), the basic principles of learning theory (the influence of antecedents and consequences on child behaviour), positive bedtime routine, giving effective instructions, partner support strategies and extinction techniques. Different types of extinction were explained to parents: standard extinction, gradual ignoring and ignoring with parental presence. They were given a choice of which to use: all chose standard extinction which was also the therapist's preference. Standard extinction involved explaining the rules to the child and after putting the child to be leaving the room and ignoring all crying or calling out. If the child came out of their room the parents were instructed to take the child immediately back to bed with minimum contact with child. If the child complied the child received positive reinforcement in the morning. Parents were advised of the possibility of an extinction burst. Modelling and role-playing was used during the sessions and written information and parent checklists also provided. Five weeks after the training ended there was a review session where goals were re-evaluated and there was training in phasing out of reinforcers. **Duration:** A minimum of 7 weeks		

If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet): In addition to the initial interview and functional assessment (conducted at the university psychology clinic) parents received three weekly training sessions and a review session (details above). The sessions on goal setting and extinction were conducted in each family home and the sessions on effective instructions and the review session were conducted at the clinic. The therapist made weekly telephone contact with parents throughout the intervention and there was daily telephone contact during the initial days of implementing extinction. Parents were encouraged to contact the therapist if they had any problems or questions. The purpose of the contact was to check progress, obtain data, answer questions, assist with problems, prompt appropriate behaviour and praise success. After the review session, contact was gradually reduced.

Description of comparator: No comparator

The outcomes measures

Outcome 1: Overall change in sleep behaviours

Details of measurement: Data for each child were displayed on graphs (based on data from sleep diaries) to allow comparison between baseline, end of treatment and 3 and 12 months follow-up. Two clinicians (one not involved with the intervention) independently visually analysed the graphs and assessed the extent of change for each child (substantial improvement, moderate improvement, no change, moderate deterioration, substantial deterioration). Definitions were provided for each of these descriptors and the raters were blinded to which sleep variable they were assessing. Where there was disagreement, raters discussed and reached consensus on a rating. For overall change in sleep behaviours the two raters agreed on 80% of the comparisons.

Outcome 2: Bedtime disturbances (per week)

Details of measurement: Defined as any disruption between being put to bed and sleep onset (e.g. calling out, leaving bedroom). Measured as above.

Outcome 3: Falling asleep in own bed

Details of measurement: Defined as number of nights per week falling asleep in own bed. Measured as above.

Outcome 4: Sleep latency

Details of measurement: The average time (minutes) between being put to bed and falling asleep. Measured as above.

Outcome 5: Night waking

Details of measurement: Number of night wakings per week that parents were aware of. Measured as above.

Outcome 6: Co-sleeping

Details of measurement: Number of nights per week child co-slept (excluding the period of falling asleep). Measured as above.

Outcome 7: Sleep duration

Details of measurement: Average duration (minutes) of sleep per week. Measured as above.

Outcome 8: Program Evaluation Questionnaire

Details of measurement: A modified version of Griffin and Hudson (1978) questionnaire. Consisted of three open-ended questions about what they liked best, least and what they would change. A fourth question asked if their child currently had a sleep problem and to rate the severity. Five items were rated on a 5-point Likert scale: parental stress levels, approval of techniques, improvement in child's sleep and behaviour, and how strongly they would recommend the programme to a friend. The final three were combined to give an overall measure of parental

satisfaction (maximum score 15).

Outcome 9: Goal Achievement Scale

Details of measurement: At the beginning of the programme parents identified two to three goals they wished to achieve in relation to their child's sleeping problem. They identified what they would consider total (100%) success for each goal. The level of success was assessed based on the sleep diaries.

Length of follow-up: End of treatment (last 4 weeks of intervention), three months after the review session and at 12 months for the children with autism.

Summary of the results:

- Overall change in sleep behaviours – *Baseline v end of intervention* (64 comparisons): substantial deterioration 0%, moderate deterioration 4.5%, no change 25%, moderate improvement 29.7%, substantial improvement 40.6%. *Baseline v 3-month follow-up* (63 comparisons): substantial deterioration 1.6%, moderate deterioration 4.8%, no change 27%, moderate improvement 23.8%, substantial improvement 41.3%. *Baseline v 12-month follow-up* (26 comparisons): substantial deterioration 0%, moderate deterioration 7.7%, no change 19.2%, moderate improvement 26.9%, substantial improvement 46.2%.
- Bedtime disturbances – For all cases the frequency of bedtime disturbances was rated as improved from baseline to end of treatment, 3 month and 12 month follow-up.
- Falling asleep in own bed – Rated as improved for 8 children from baseline to end of treatment, though one child had shown a trend towards improvement during the baseline period. Seven maintained the improvement at both follow-ups. Improvement was not expected for two children as this was not a problem at baseline.
- Sleep latency – Rated as improved for 6 children from baseline to end of treatment, though one child had shown a trend towards improvement during the baseline period. Two children were rated as deteriorated and 2 as unchanged. Five maintained the improvement at follow-up but one deteriorated.
- Night waking – Rated as improved for 7 children at the end of intervention and at follow-up, though one child had shown a trend towards improvement during the baseline period. Three children were rated as unchanged though change was not expected for 2 as this was not a problem at baseline.
- Co-sleeping – Of the 6 children for whom this was a problem at baseline this was rated as improved at end of intervention, at 3 month follow-up and for 5 at 12 month follow-up.
- Sleep duration – The authors state that there was little consistency among participants in the rate of change across time.
- Parents views of sleep problem (from Program Evaluation Questionnaire) – Five of the ten mothers stated that their child still had a sleep problem after the intervention. In four of these cases the severity had decreased.
- Goal Achievement Scale – At end of intervention 12 out of 25 goals were achieved with 100% success and the mean Goal Achievement Score was 76.3%. In the autism group there was further improvement at 3-month follow-up (mean GAS 80.8) and at 12 months (mean GAS 89%). For the FXS group at 3-months the level of achievement increased for 4 goals and decreased for 4.

Any negative consequences: Seven participants experienced an extinction burst in the week that extinction was implemented.

Views of parents: Program Evaluation Questionnaire – parents said the best aspects of the program were the outcome, the support provided, and the method of training. Record keeping was

the thing they liked least. Two found it difficult to stick to a bedtime routine, one found the training sessions too long, three thought the programme time consuming. The mean parental satisfaction score was 13.8 (range 11 to 15). All said they would recommend the programme to a friend.
Authors' conclusion: The results support the hypothesis sleep problems of children with autism or FXS will reduce after behavioural intervention.
Comments: The authors raise a number of points to consider when interpreting the findings. 1) Extinction did not seem appropriate for early morning waking or night rocking possibly because they were not positively reinforced by parental responses prior to treatment. 2) They observe that in most cases improvement did not occur until extinction was implemented. 3) The extent to which the findings can be applied to a wider population is limited as the intervention needs to be tested across a wider range of disabilities. 4) They point out that the two children that were withdrawn from the study were more non-compliant than those who remained and were also older. They suggest that extinction may be too difficult or stressful to implement with extremely non-compliant or older children.

Publication details		
Author: Wiggs[16]	**Year:** 1998	**Related publications:** Wiggs[33] Wiggs[34]
Stated aim: To explore the efficacy and mechanisms of treatment in children with severe learning disabilities, severe sleep problems and severe daytime challenging behaviour		
Study design: RCT (schools rather than families were randomly allocated to intervention or control in order to avoid discussion of the intervention between parents in the two groups)		
The participants		
Number: N=31	**Age:** Intervention (n=15) – mean 8.21yrs (SD 2.7); Control(n=15) – mean 10.77yrs (SD 3.81)	**Sex:** 18 males, 12 females
Type of disability: The children had severe learning disabilities (Down syndrome, meningitis, microcephaly, cerebral palsy, CHARGE association, agenesis of the corpus callosum, Sanfillipo syndrome, Ring 15 chromosome disorder and unknown with autism). Eleven children also had uncontrolled epilepsy.		
Sleep problem: 10 settling; 6 settling and night waking; 5 settling, night waking and sleeping in parental bed; 1 night waking, 2 settling and sleeping in parental bed; 2 night waking and early waking; 2 night waking and sleeping in parental bed; 1 settling, night waking and sleeping in parental bed. For entry into the study children had to have a severe sleep problem (based on specific criteria). **How the sleeping problem was assessed:** Based on a detailed sleep history using a semi-structured interview. A severe sleep problem was defined as settling problems of more than one hour duration 3 or more times per week or night waking 3 or more times per week where the child disturbed parents or went into parents room or early waking before 5am, 3 or more times per week. **Other information:** Children were eligible for the study if they had a severe sleep problem and one or more daytime challenging behaviours (any item assessing challenging behaviour on the Aberrant Behaviour Checklist classified as quite serious or severe). They were recruited from families who had responded to a survey of special schools. There were 486 children included in they survey of whom 209 families completed a questionnaire (43%). 51 children met the inclusion criteria for the sleep study of whom 31 agreed to participate. One dropped out from the intervention group before it commenced. Of the 20 who declined 10 were too busy, 7 said their child's sleep had improved and the reason was unknown for 3.		
The intervention		
Setting: Home-based **Type of behavioural intervention:** A range of behavioural techniques depending on the problem and parent preferences **Description of intervention:** Following a preliminary introductory visit to explain baseline questionnaires and the activity monitor watch there was a 1.5 to 2.5 hour visit to undertake a functional analysis of the problem. For the intervention group, a detailed behavioural programme was agreed. There was discussion of possible mechanisms maintaining sleep problems and the advantages and disadvantages of different approaches such as extinction, graded extinction, stimulus control procedures and positive reinforcement. Parents' aims for treatment and target(s) for the first stage were identified. After this visit parents were sent a written outline of the agreed behavioural programme. **Duration:** One month		

If delivered by parents, give description of training and support received (including methods of delivery of support to parents for the intervention (e.g. face to face, telephone, booklet):
In addition to the visit where the intervention was delivered progress was monitored by regular telephone calls. Both the intervention and control group received the preliminary visit and four visits to deliver and collect questionnaires.
Description of comparator: Waiting list control

The outcomes measures

Outcome 1: Composite Sleep Index
Details of measurement: Modification of the Simonds and Parraga Sleep Questionnaire (1982). Scores frequency and duration of settling and night waking problems and frequency of early waking and sleeping in parental bed. Possible score range from 0 (no problem) to 12.
Outcome 2: Activity monitor (child and mother)
Details of measurement: The wrist watches were worn for three nights at each assessment period by the child and mother. Movement was calculated for every 30seconds during the recording period. Sleep period (time from sleep onset to waking), activity score (mean value of movement during sleep), movement index (% of sleep period spent moving) and fragmentation index (% of immobile phases during sleep period which were 30 seconds duration or less) were measured.
Outcome 3: General daytime behaviour
Details of measurement: 18 items enquiring about challenging behaviour from the Aberrant Behaviour Checklist (ABC) (Aman & Singh 1986) which were rated by mothers and teachers (baseline and 3 month follow-up only). These were entered into a factor analysis and five distinct categories of behaviour identified: irritability, lethargy, stereotypies, hyperactivity and inappropriate speech.
Outcome 4: Severity of challenging behaviour
Details of measurement: Mean severity rating by mother and teachers of each of 5 challenging behaviours: aggression, non-compliance, self-injury, temper tantrums and screaming.
Outcome 5: Frequency of challenging behaviour
Details of measurement: Mean severity rating by mother and teachers of each of the 5 challenging behaviours.
Outcome 6: Parental satisfaction with sleep
Details of measurement: Rated satisfaction with their own sleep and satisfaction with their ability to cope with their child's sleep pattern and daytime behaviour on a 6-point Likert scale from 0 totally satisfied to 6 totally unsatisfied.
Outcome 7: The Malaise Inventory (Rutter, Tizard & Whitmore 1970)
Details of measurement: 24-item binary choice questionnaire to assess parental stress. Test-retest reliability reported to be high.
Outcome 8: Epworth Sleepiness Scale (Johns 1991)
Details of measurement: 8-item self-report scale assessing daytime sleepiness. The items assess likelihood of falling asleep in everyday situations. Possible score ranges from 0 to 24 (maximum sleepiness).
Outcome 9: Internal/External Locus of Control Scale (Rotter 1966)
Details of measurement: 29-item forced choice to measure orientation to internal or external control beliefs.
Outcome 10: Perceived control
Details of measurement: Parents rated their ability to control any sleep-related problems shown

by their child on a 100mm visual analogue scale with higher score indicating greater perceived control.

Length of follow-up: One month and 3 months following commencement of treatment.

Summary of the results:

- Composite Sleep Index – Based on 2x3 ANOVA there was a statistically significant main effect for time ($p<0.001$), group ($p=0.001$) and a significant interaction between group and time ($p<0.011$). Based on post-hoc tests (Scheffe's test) there was a statistically significant improvement from baseline to one month and baseline to 3 month follow-up for the intervention group: mean 6.73 (SD 2.31); 3.79 (SD 1.89) and 2.96 (SD 2.24) respectively; but no change for the control group mean 7.23 (SD 2.26); 6.62 (SD 1.89) and 6.29 (SD 2.70) respectively.
- Activity monitor – *Children's movements*: There were no between group differences. There was a statistically significant main effect for time only on each of the sleep variables. Based on post-hoc tests there was an improvement for both groups from baseline to 1 and 3-month follow-up for sleep period, activity score and movement index and improvement from baseline to 1-month for the fragmentation index but deterioration between 1 and 3-month follow-up. *Mothers' movements* – There was a statistically significant interaction between group and time ($p=0.03$) for sleep period. Based on post-hoc tests mothers in the intervention group showed an increased sleep period between baseline and 1-month and 3 month follow-up. There was a statistically significant main effect for time for the movement index ($p=0.011$). Based on post-hoc tests the intervention and control group showed a significant improvement from baseline to 1-month follow-up.
- General daytime behaviour – There were no statistically significant differences between intervention and control in how they changed over time. There was a statistically significant decrease in both groups from baseline to 1 and 3 month follow-up in irritability, lethargy and hyperactivity based on mother's ratings and for irritability and hyperactivity from baseline to 3 months on teachers rating.
- Severity of challenging behaviour – There were no statistically significant differences between intervention and control over time for mother and teacher ratings. Based on teacher ratings aggression, temper tantrums and noncompliance significantly decreased over time in both groups and noncompliance based on mother ratings.
- Frequency of challenging behaviours – There were no statistically significant differences between intervention and control over time for mother and teacher ratings. Based on teacher ratings there was a significant decrease in frequency of challenging behaviours over time in both groups.
- Parental satisfaction with sleep – *Mothers* (n=15 for each group): there was a statistically significant group by time interaction for satisfaction with own sleep, satisfaction with child's sleep and satisfaction coping with child's sleep. There was improvement from baseline to 1-month and 3-month follow-up which was greater in the intervention group. *Fathers* (12 in treatment group and 13 in control group): there was a statistically significant group by time interaction for satisfaction with own sleep, satisfaction with child's sleep. There was improvement from baseline to 1-month and 3-month follow-up which was greater in the intervention group.
- The Malaise Inventory – *Mothers*: there was a statistically significant group by time interaction for stress ($p=.053$). Mothers in the intervention group reported reduced stress from baseline to 3-month follow-up. *Fathers*: there were no statistically significant between

group differences over time.

- Internal/External locus of control – *Mothers*: there were no statistically significant between group differences over time. *Fathers*: there was a statistically significant group by time interaction for externality. There was an increase post-intervention for the intervention group and a reduction for the control group.
- Perceived control - There were no statistically significant differences between intervention and control over time amongst mothers or fathers.

Any negative consequences: None reported

Views of parents: None reported

Authors' conclusion: Sleep problems can be successfully treated in this group of children but the mechanisms of treatment may not be as direct as supposed. The intervention did not appear to be associated with any change in the children's daytime behaviour. Such interventions can have a significant positive impact upon mothers, and to a lesser degree, fathers. There was evidence of improvement over time in child and parent outcomes for both the intervention and control group suggesting nonspecific effects of participating in the study.