1. Title of the article: **Long-term impact of techniques used to manage sleep disturbance in under-5s: a rapid systematic review**
2. Please find below the names of the authors, affiliations and street addresses:

Gilly Mancza\* and Dr Wendy Wigleyb

aSenior Teaching Fellow, School of Health Sciences, University of Southampton, Southampton, United Kingdom; bInterim Head of School, School of Human and Social Science, University of West London, Ealing, United Kingdom

1. Author Responsible for Correspondence: Gilly Mancz,

Address: University of Southampton, School of Health Sciences, Building 67, University Road, Highfield Campus, Southampton, SO17 1BJ, United Kingdom. Email: g.j.mancz@soton.ac.uk Telephone number: 02380 598612 Twitter: @gillyjm1

**Long-term impact of techniques used to manage sleep disturbance in under-5s: a rapid systematic review**

**Abstract**

Behavioural insomnia is common in under-fives and modified extinction interventions are the recommended treatment. There is contention regarding whether techniques that promote ignoring a child’s distress causes potential damage to infant mental health and the mother-child bond. A rapid review of the literature was undertaken to explore the association between use of behavioural interventions and infant mental health, and/or maternal mental health and/or parent-child attachment. Three themes were identified from the literature; (1) the impact of behavioural interventions on infant and maternal mental health, (2) behavioural interventions and maternal mood and/or stress, (3)J parental perceptions of behavioural interventions on their child and themselves. The key recommendations and implications for practice are explored and clarified.

*Keywords*: infant, emotional, health visitors, mother/s, sleep disturbance

## Introduction

Sleep disturbances in childhood are commonplace (Moturi & Avis, 2010; Gregory & Sadeh, 2012; Vriend et al, 2013; Bathory & Tomopoulos, 2017), affecting approximately 25% of all children (Owens, 2008), and encompass a range of disorders with both organic and behavioural causes (Mindell & Owens, 2009). The most frequently reported sleep disturbances in early childhood are behavioural in nature and consist of either sleep onset association disorder, limit setting disorder or a combination of both disorders (Hill, 2011). Infant sleep problems are often a major cause of concern for parents (Mindell et al, 2010) and it is recognised that there are a number of associated detrimental outcomes for young children who experience sleep deprivation, such as an impact on cognitive development, attention skills, mood, behaviour and health (Mindell et al, 2012; Heussler et al., 2013).

Negative impacts have been found to be associated with moderate sleep reductions, for example the pre-school age group have an increased risk of becoming obese if they slept for less than twelve hours as an infant (Taveras et al, 2008).  Furthermore, the evidence suggests there is a risk that sleep disturbances that present in babies can develop into chronic sleep problems in childhood and adolescence (Kuhn & Weidinger, 2000; Lam et al, 2003; Mindell et al., 2006; Blunden, 2011). On-going and chronic sleep issues can also have a long-term negative impact on the wider family, such as a significant increase in mothers developing postnatal depression (PND), family breakdown, poor health and, in rare cases, result in child abuse (Bayer et al, 2007; Martin et al, 2007; Smart & Hiscock, 2007; Giallo et al, 2011).

Behavioural insomnias are influenced by a complex interplay between environmental, biological, developmental and cultural factors (Mindell et al., 2010; Moturi &Avis, 2010).  Behavioural interventions are the recommended treatment for these types of insomnia and are predominantly based on extinction or modified extinction strategies which encourage parents to withdraw and ignore their child’s unwanted behaviour (Morgenthaler et al., 2006; Hill, 2011).  The aim of these types of strategy is to assist young children in learning to self-sooth whilst improving sleeping patterns and parental well-being (Sadeh et al, 2011). However, advances in neuroscience have resulted in a growing understanding of brain development and the influence of experiences during the the antenatal period and early childhood, the childhood environment and attachment to a primary caregiver (Shore, 2003; Gerhardt, 2004; UNICEF, 2014; Newman et al, 2015).  Babies are born with an innate need to seek comfort and parental responsiveness can determine whether the baby is supported to regulate their emotions, stress response and level of neurophysiological arousal (Gerhardt 2004, Bernard & Dozier, 2010; Newman et al, 2015). An unregulated stress response, such as in response to being left to cry or feelings of abandonment, can be toxic to the developing brain, affecting key structures such as the hypothalamus and prefrontal cortex as well as other areas of the body, such as the immune system (Gerhardt, 2004). This has led some professionals to question the use of behavioural interventions in light of the potential impact the stress response may have on the developing brain and parent-child dyad.

The on-going contention regarding whether behavioural interventions cause harm is polarised and often emotionally charged within the literature (Sadeh et al., 2011).  Advocates of behavioural interventions highlight that pure extinction techniques are rarely recommended, in contrast best practice guidelines such as modified extinction strategies do promote degrees of parental responsiveness, have not been found to cause harm and are linked to positive outcomes, including improved maternal mood (Sadeh et al., 2011; Price et al, 2012).  Whereas opponents of extinction based strategies argue that an unregulated stress response can be toxic to the developing brain, emotional health and furthermore negatively impact on the parent-child bond (Blunden et al, 2011). Expert bodies also provide conflicting advice; the American Academy of Pediatrics (2012) and the UK’s National Health Service (NHS) (NHS Choices, 2012; 2016) advise behavioural interventions are safe and improve sleep, whereas the Australian Association for Infant Mental Health (AAIMH) (2013) advise against the use of such techniques due to the risk to infant mental and emotional health.  This discord has filtered through to parental experience and how to manage sleep is acknowledged to be one of the most contentious issues to discuss on parenting blogs and online peer support groups, often resulting in the expression of strong views and judgements (Strange et al, 2018). Parents, and the health professionals supporting them, lack confidence in managing sleep and experts sitting on both sides of the debate have called for further research to be undertaken to resolve whether behavioural interventions cause harm (Blunden et al., 2011; Sadeh et al., 2011).

This rapid systematic review aimed to investigate the existing literature that specifically examined associations between the use of behavioural interventions and infant mental health, and/or maternal mental health and/or parent-child attachment.

## Method

### Study design

To allow for a concise analysis of the literature and to inform professional practice regarding supporting parents with infant sleep disturbances, a rapid review of the literature was undertaken (Khangura et al, 2012), following the Preferred Reporting Items for Systematic Review and Meta-Analyses (PRISMA) statement (Moher et al, 2009).  The following recognised modifications were employed to allow for a rapid rather than full review; (1) a reduced list of sources were searched, (2) only articles published in English were considered, (3) only studies published in the last 10 years were included, (4) a range of search tools were employed and (5) only one reviewer undertook the data extraction (Haby et al., 2016).

### Inclusion criteria

Articles were included in the rapid review if they met the inclusion criteria; (1) published in English, (2) published between January 2007 and June 2018 as this insured currency, (3) studies undertaking primary research into behavioural techniques and their outcomes on infant mental health and/or maternal mental health and/or parent child attachment, (4) studies undertaken in westernised countries as there are recognised cultural factors that influence sleep, (5) focus on the under-five age group as this is the most common age range to experience behavioural interventions and key age for brain development, and (6) studies undertaken on normally developing children as children with additional needs often have further complex factors influencing their sleep.  Both quantitative and qualitative research methods were to be included in the review, as this would have broadened the research to include lived experience of implementing a sleep intervention, however, only research with a quantitative focus was identified.

### Search methods

An extensive search of three databases and two websites was undertaken.  The three databases searched were: Cumulative Index to Nursing and Allied Health Literature, MEDLINE and PsycINFO.  The two websites searched were Google and Google Scholar – two studies were identified via Google Scholar. To minimise the risk of bias, a search of grey literature, including doctoral dissertations, was conducted and a hand search of a peer reviewed journal (Journal of Health Visiting, 2018) that does not feature in the databases but focuses on health visiting practice in the UK was included.   No studies were identified through the search of the grey literature or hand search of the journal. A reference list search was conducted and one further study was identified.

### Search strategy

The search of the databases was undertaken in May 2018 and the search of the websites, grey literature, hand searching and reference screen was undertaken in June 2018.  The search terms were separated into three areas; Population (part 1) terms used were: “babies” OR “infant\*” OR “toddler\*” OR “pre-schooler\*” OR “child” OR “child” (with age cap using the database limits within the search engine) OR “parent\*” OR “carer\*” OR “mother”; Population (part 2) terms used were ”sleep\*” OR “Insomnia\*” OR “waking\*”; Intervention terms used were “extinction” OR “graduated extinction” OR “modified extinction” OR “controlled cry\*” OR “cry\*” OR “gradual withdrawal” or “gradual retreat” OR “modified extinction with parental presence” OR “self-sooth\*”.   The final stage involved combining the results for Population (part 1), Population (part 2) and Intervention using the logical AND operator. The internet search used the following terms: “controlled crying”; “infant sleep and sleep training”; “behavioural sleep intervention and attachment”. Duplicate papers were excluded.

### Screening and study selection

The searches of the databases and hand searching was undertaken by one author (GM) and search of the websites and grey literature was undertaken by both authors (GM & WW).  The titles and abstracts were reviewed against the inclusion criteria by one author (GM) – any titles and abstracts that potentially met the inclusion criteria were included for the final review and the full papers were retrieved.  The papers were then reviewed against the inclusion criteria by one author (GM) and the final papers were identified. The final papers were discussed with the second author (WW) and agreed.

### Data Extraction

The following data was extracted from each study; country of study, study design, sample characteristics (age of children, socioeconomic background, level of parental education, ethnicity), sample size, study methodology, results and findings.  To determine the reliability of the study design and methodology and to judge the risk of potential bias, each paper was systematically reviewed using the Bluff and Cluett Framework (Bluff & Cluett, 2006).

Full-text articles excluded, with reasons  
(n = 7)

Not primary research (commentaries and reviews)= 7

Studies included in quantitative synthesis

(n = 10)

Records excluded  
(n = 82)

Full-text articles assessed for eligibility  
(n = 17)

Records screened  
(n = 99)

Records after duplicates removed  
(n = 99)

Identification

## IdentificationIden

Records identified through database searching (n = 131)

Additional records identified through other sources  
(n = 3)

## SScSc

Screening

Eligibility

## Eligibility

Included

## Included

##### Figure 1: Flowchart of Study Selection

## Results

### Search Results

In total, 134 studies were retrieved through the search, with ten studies being eligible for review, see figure 1. Five studies focused on the impact of behavioural interventions on infant and maternal mental health, three studies analysed behavioural interventions and maternal mood and/or stress and two studies explored parental perceptions of behavioural interventions on their child and themselves.

### Study characteristics

Table 1 provides an overview of the five studies focused on the impact of behavioural interventions on infant and maternal mental health (Hiscock et al, 2008; Matthey & Črnčec, 2012; Middlemiss et al, 2012; Price et al., 2012; Gradisar et al., 2016). Table 2 gives an analysis of the impact of behavioural interventions on maternal mood and/or stress (Wade et al, 2007; Blunden, 2011; Hall et al., 2015).  Table 3 provides a summary of the two papers concentrating on the parental perceptions of behavioural interventions on themselves and their infant (Loutzenhiser et al, 2014; Honaker et al, 2018). All of the papers were primary studies involving parents of children aged under-five years, experiencing sleep difficulties. Of the ten studies included in the review, all were based in westernised countries; five were conducted in Australia, two in Canada, two in the USA and one in New Zealand.  Each of the ten studies were quantitative in approach; four were randomised controlled trials, two survey designs, one time series design, one a single case replication design, one group design and one group only design. Tables 1, 2 & 3 expand on the validated and non-validated tools used to collect data and measure variables.

| **Study title, Author(s) and Country** | **Study Aims** | **Population and Sample size** | **Data Collection** | **Data Analysis** | **Findings** | **Implications for practice** |
| --- | --- | --- | --- | --- | --- | --- |
| *Long-term Mother and Child Mental Health Effects of a Population-Based Infant Sleep Intervention: Cluster-Randomised, Controlled Trial*  Hiscock et al (2008)  Australia | To analyse the long-term effects of a behavioural intervention on infant mental health and sleeping patterns, maternal mental health and parenting style. | Parents of infants aged 8-10 months reporting a sleep problem when their child was 7 months old to be included in the study.  328 families. | Quantitative approach: cluster randomised controlled trial.  Questionnaires and structured consultations.  Validated tools: EPDS, Parent Behaviour Checklist, Infant Behaviour Checklist, Global Infant Temperance Scale.  Unvalidated tools: questionnaire including questions regarding sleep. | Descriptive and inferential statistical tests  Descriptive statistics; mean, standard deviation, confidence interval and odds ratio.  Inferential statistics; *p-*value of probability and significance.  Data was presented in tables and provided a comparison between the control and intervention groups. | Mothers in the intervention group were less likely to be clinically depressed when their infant was two years.  Neither parenting styles nor infant mental health were markedly different between the control and intervention group.  Mothers with poor educational levels and from deprived backgrounds dropped out at a higher rate than other socioeconomic groups. | First randomised trial to analyse the long-term effects of behavioural interventions for infant sleep disturbances.  Behavioural intervention had a significant impact in reducing maternal depression over the two years, whilst having no long term positive or negative impact on parenting practices or child mental health.  Well child systems could deliver behavioural interventions at a population level to reduce negative impacts associated with poor infant sleep. |
| *Comparison of two strategies to improve infant sleep problems, and associated impacts on maternal experience, mood and infant emotional health: A single case replication design study*  Matthey S and Črnčec R (2012)  Australia | To compare the outcomes of two behavioural interventions on infant sleep, infant mental health and maternal mood. | Parents with infants aged 6-18 months who sought professional support for sleep disturbances.  16 families. | Quantitative approach: single case replication study design.  Sleep diaries and questionnaires.  Validated tools: EPDS, Hospital Anxiety and Depression Scale, and the Experience of Motherhood Questionnaire.  Unvalidated tools: Infant emotional health questionnaire, sleep diaries. | Descriptive and inferential statistical tests.  Descriptive statistic; mean.  Inferential statistics; Cohen *d*, tests of significance and a permutation test.  Data was presented in tables and graphs and showed the pre and post test results. | Both controlled crying and modified extinction with parental presence were successful at treating the majority of infant sleep disturbances.  No reported negative emotional or behavioural impacts associated with either behavioural intervention on any infant participating in the study, instead an overall improvement was found in infant behaviour and emotional health.  Behavioural interventions were not associated with negative outcomes for young children. | First study to specifically focus on the impact of two interventions, controlled crying and modified extinction with parental presence, on maternal and infant mental health.  Both interventions were found to be effective at resolving sleep disturbance and neither intervention was found to have a negative impact on infant mental health, with mothers in both groups reporting improvements to their mood and experience of motherhood.  Findings relevant to health professionals supporting families with poor infant sleep as these are the commonly recommended treatments for sleep disturbance, and provide further evidence for offering alternative interventions, if appropriate. |
| *Asynchrony of mother-infant hypothalamic-pituitary-adrenal axis activity following extinction of infant crying responses induced during the transition to sleep*  Middlemiss et al (2012)  New Zealand | To examine mother and child synchrony when undertaking a 5 day behavioural sleep programme in a residential unit. | Mothers with infants aged 4-10 months referred to a residential hospital in the Northern Y District of New Zealand for intensive sleep support.  All families on the waiting list were invited to take part.  The exclusion criteria prevented mothers who smoked, took antidepressants or anti- inflammatory steroid medications from participating as these all effect cortisol production.  A total of 25 mother-child dyads took part in the study. | Quantitative approach: one-group-only-design.  Mother and child salivary samples, documented observations of the infant’s signalling behaviour at bedtime and maternal questionnaires.  The study used several unvalidated tools. | Descriptive, inferential and correlation statistical tests.  Descriptive statistics; mean and standard deviation.  Inferential statistics: *t-*tests and tests of significance.  Correlation statistics: Pearson’s correlation test.  The data was discussed but was not presented visually. | The study found that on day one the mothers’ and their babies’ behavioural and physiological responses to the extinction sleep programme were synchronised, however, by day three the babies had stopped demonstrating behavioural signs of distress but their physiological response remained high, whereas the mother’s behavioural and physiological response lowered causing asynchrony. | First study to examine the physiological impact on infants when conducting a behavioural sleep programme.  Focus of study was mother-child synchrony rather than the impact of a sleep programmes.  Not generalisable to the UK and populations served by public health professionals.  An infant’s physiological response may be different from the behavioural cues exhibited and the potential impact on infant mental health should be considered.  Further research is required to explore the asynchrony between mother and child and the wider impact on infant development. |
| *Five-Year Follow-up of Harms and Benefits of Behavioural Infant Sleep Intervention: Randomized Trial*  Price et al 2012  Australia | To analyse the long-term effects of a behavioural intervention on infant mental health and sleeping patterns, as well as maternal mental health and attachment. | To be included in the follow up, the families had to have taken part in the original study.  225 of the eligible 326 families took part in the study. | Quantitative approach: five year follow up of a cluster randomised controlled trial.  40-60 minute assessment and cortisol samples.  Validated tools: Pediatric Quality of Life Inventory, cortisol samples.  Unvalidated tools: questionnaire. | Descriptive and inferential statistical tests.  Descriptive statistics; mean, standard deviation and confidence interval.  Inferential statistics; linear regression, *p-*value of probability and significance.  Data was presented in tables and provided a comparison between the control and intervention groups. | No difference between long-term outcomes for the intervention and control groups, including areas where previously concern has been raised regarding the impact of using sleep interventions. | First paper to follow up a large RCT study to determine the long term outcomes of using behavioural interventions when the children reach the age of 6 years.  No positive or negative long term effects on the child, child-parent relationship or maternal mental health when implementing a sleep intervention.  Health professionals can recommend the delivery of behavioural interventions to reduce negative impacts associated with poor infant sleep in the short and medium term. |
| *Behavioural Interventions for Infant Sleep Problems: A Randomized Controlled Trial*  Gradisar et al 2016  Australia | To examine the effects of behavioural interventions on infant and parent sleep, infant stress, child behaviour and parent-child attachment. | Parents with infants – age range was not specified – who identified their child as having a sleep problem and the child had previously met expected milestones and weight gain at the under 1-month check.  43 families took part in the study with infants aged between 6-16 months. | Quantitative approach: randomised controlled trial.  Questionnaires, an interview, salivary samples, sleep diaries, ankle worn activity monitor, maternal depressed mood and stress assessment and strange situation test.  Validated tools: DASS-21, actigraphy, cortisol samples, strange situation test.  Unvalidated tools: sleep diary, questionnaire and 90 minute interview regarding infant health and sleep. | Descriptive and inferential statistical tests.  Descriptive statistics; mean and confidence intervals.  Inferential statistics; Cohen *d*, tests of significance and regression.  Data was presented in tables and graphs and showed the during and post intervention test results. | Graduated extinction and bedtime fading had a significant impact on sleep compared to the control group.  The control group were offered sleep advice and it was identified that sleep advice may not be effective on its own to improve poor sleep in young children.  Cortisol was collected twice daily during the intervention period and cortisol levels were found to be within normal limits.  No differences in parent-child attachment styles or child behaviour between the control group and intervention groups were identified at the 12 month follow up. | Second study to examine the physiological impact on infants when conducting a behavioural sleep programme.  Findings would be generalisable to the UK and populations served by health professionals.  Neither of the interventions resulted in chronically elevated cortisol levels - – this is important as one of the main concerns raised regarding extinction based programmes is that they could result in chronically elevated cortisol levels causing a negative impact on parent-child attachment and child development.  Further research is required to explore the impact of different sleep based interventions. |

Table 1 -  Overview of the five studies focused on the impact of behavioural interventions on infant and maternal mental health

| **Study title, Author(s) and Country** | **Study Aims** | **Population and Sample size** | **Data Collection** | **Data Analysis** | **Findings** | **Implications for practice** |
| --- | --- | --- | --- | --- | --- | --- |
| *Two-Session Group Parent Training for Bedtime Noncompliance in Head Start Preschoolers*  Wade et al (2007)  USA | To examine the effectiveness of delivering a behavioural intervention in a group setting to low income ethnic minatory parents on sleep, as well as analysing impact on parental mood and stress. | Parents of children aged 4-5 years accessing Headstart provision.  The child had to meet the criteria for difficult bedtime behaviour.  Five families. | Quantitative approach: time series design.  Parent interviews, diaries and video recordings.  Validated tools were used; ECBI, PSI-SF and the Beck Depression Inventory-II.  Unvalidated tools including the sleep diaries were designed for the study. | Descriptive and inferential statistical tests.  Descriptive statistics; mean and standard deviation.  Inferential statistical; t-test, tests of significance and regression.  Data was presented in tables and graphs. Comparisons were made before and after the intervention. | Delivery of a sleep programme (controlled crying) in a two session group setting was effective at improving sleep in low income, ethnic minority children.  Effectiveness similar to that of the existing evidence base.  Parental stress and depression levels were found to reduce post intervention and child daytime behaviour improved. | First paper to specifically analyse behavioural interventions in a vulnerable population and to evaluate the effectiveness of a group programme.  Small size of the study, payment and daily contact impacts on overall generalisability.  Further research needed. |
| *Behavioural treatments to encourage solo sleeping in pre-school children: An alternative to controlled crying*  Blunden S (2011)  Australia | To investigate the impact of a no cry technique on infant sleeping patterns and maternal quality of life. | Parents with children aged under five years who attended a private sleep clinic.  33 families. | Quantitative approach: one group design.  Two questionnaires.  Study used unvalidated tools. | Descriptive and inferential statistical tests.  Descriptive statistics; mean, standard deviation and range.  Inferential statistics; Cohen’s *d* and the *p-*value of probability and significance.  Data was presented in tables showing the differences in data pre and post intervention. | A modified behavioural technique which did not require parents to ignore their child’s distress resulted in a significant improvement in sleep patterns and reduced negative sleep associations and co-sleeping.  Parent satisfaction with the technique was found to be high. | First study to address the difficulties parents have in ignoring their child’s distress when implementing a sleep programme through the creation of a modified behavioural technique.  Findings from the study are not generalisable to wider public health professionals working in the UK.  Further research is recommended to inform the evidence base and build on the findings. |
| *A randomised controlled trial of an intervention for infants’ behavioural sleep problems*  Hall et al (2015)  Canada | To determine the effects of an intervention on infant sleep and parental mood. | Parents of infants who were aged 5½ - 8 months old that met the clinical classification of a sleep onset association disorder.  235 families. | Quantitative approach: randomised control trial.  Data collected using questionnaires, sleep diaries and actigraphy.  Validated tools were used; MAF, PSQI and MCISQ.  Unvalidated tools: the sleep diaries were designed for the study. | Descriptive and inferential statistical tests.  Descriptive statistics; mean and standard deviation.  Inferential statistics; the two-tailed *p-*value of probability, Fisher's exact test, confidence intervals and Mantel-Haenszel test.  Data was presented in tables showing the differences in data between the intervention and control groups. | Intervention and control group received their information through a group setting, with follow up telephone calls.  Primary and secondary care givers in the intervention group were found to have improved sleep quality, decreased levels of fatigue and increased confidence at managing their child’s sleep issue compared to the control group.  Mood improved and feelings of anger regarding their infant’s sleep issue decreased for the primary caregivers in the intervention group compared to the control group.  A significant difference was not found between secondary caregivers. | Findings from this study would be generalisable to the UK and populations served by public health professionals.  The intervention was successfully delivered using a group format.  Further research is required to explore the impact of different sleep based interventions on attachment to analyse the impact on the infant. |

Table 2 - Summary of the three studies which analysed behavioural interventions and maternal mood and/or stress

| **Study title, Author(s) and Country** | **Study Aims** | **Population and Sample size** | **Data Collection** | **Data Analysis** | **Findings** | **Implications for practice** |
| --- | --- | --- | --- | --- | --- | --- |
| *Parental perceptions of the effectiveness of graduated extinction in reducing infant night wakings*  Loutzenhiser et al (2014)  Canada | To examine Canadian parents’ perception of using the controlled crying technique in the community. | Parents of infants aged 6-18 months old.  411 families. | Quantitative approach: internet survey.  On-line questionnaire.  Study used an unvalidated data collection tool. | Descriptive and inferential statistical tests.  Descriptive statistics; mean and standard deviation.  Inferential statistics; regression and the *p-*value test of significance.  The data was discussed within three different sections and the results were presented in tables which showed the frequency of the variables and correlations between the variables. | Over 50% of the participants were using the controlled crying technique, however, only half of the families who had used the intervention had found it successful in reducing night wakings.  Parental feedback showed a strong association with using the technique and parental and infant stress, although the reasoning for this was not explored within the survey.  A relationship was identified between family support and stress levels – those with good support reported lower levels of stress when implementing the technique. | First paper to conduct a large scale survey regarding parental experiences of using controlled crying in the community.  Generalisability reduced as the sample mainly consisted of well-educated Caucasian women.  Many of the parents that professionals come into contact with may have already tried controlled crying without success and this may impact on their willingness to put the strategy into place again.  Further research is advocated to explore parental experience of using behavioural techniques. |
| *Real-World Implementation of Infant Behavioural Sleep Interventions: Results of a Parental Survey*  Honaker et al (2018)  USA | To investigate parental perception of using behavioural sleep interventions in the community. | Parents of infants or toddlers (age range not specified) who had joined a Facebook peer support group for parents using or intending to use behavioural sleep interventions.  652 families. | Quantitative approach: internet survey.  On-line questionnaire.  Questionnaire used validated questions from the Brief Infant Sleep Questionnaire and other non-validated tools. | Descriptive and inferential statistical tests.  Descriptive statistics; mean, mode, standard deviation and confidence intervals.  Inferential statistics; chi square test and anova.  Data was presented in tables and graphs, showing overall results and differences between different behavioural interventions. | The majority of parents successfully reported implementing one of four sleep interventions in the community – unmodified extinction, modified extinction, parental fading or parental fading with support and all four types of intervention were reported to improve their child’s sleeping patterns.  Unmodified extinction was found to have a higher first-time success rate for implementation than unmodified extinction, whereas parental presence with support had the lowest parental satisfaction rating.  The majority of participants were found to be using extinction based strategies. | Provides insight into parental experience of behavioural techniques within the community, particularly around parental stress when implementing techniques and length of infant crying.  These may be helpful for health professionals to be aware of when offering guidance to parents, particularly around expectation setting.  The majority of parents who completed the questionnaire were white and educated, reducing the generalisability of the findings.  The paper highlights the majority of parents are commencing sleep programmes at a young age, often before 6 months, and this may have wider implications on night feeds and growth.  Further research is recommended to examine the optimum age and/or weight an infant can safely stop night feeds without adverse effects. |

Table 3 - Analysis of the two studies exploring parental perceptions of behavioural interventions on their child and themselves.

### Ethical Considerations and risk of bias

Each of the studies were critiqued using the Bluff and Cluett Framework (Bluff & Cluett, 2006), which analyses study design, method, ethical considerations, bias, data collection, data and findings to determine the overall validity of the study and implications for practice.

When critiquing ethical considerations eight out of the ten papers provided details of their ethical approval and consent processes (Hiscock et al., 2008; Matthey & Črnčec, 2012; Middlemiss et al, 2012; Price et al, 2012; Loutzenhiser et al, 2014; Hall et al, 2015; Gradisar et al, 2016; Honker et al, 2018), whereas two studies did not provide any information regarding ethical approval, although a consent process was evident during the methods section (Wade et al, 2007; Blunden, 2011).  The lack of information regarding ethical approval and oversight was concerning in one study, as it focused on low income and minority families, offering them both free treatment and a US$150 payment for taking part, with one participant excluded as they were found to be providing fictional accounts of their child’s sleep patterns (Wade et al, 2007).

The ethical dilemma of withholding sleep support and hence the choice of a quasi-experimental study design rather than a RCT was cited in three out of the ten studies (Wade et al, 2007; Blunden, 2011; Matthey & Črnčec 2012). The challenge of blinding participants to the aims of the study when they were aware of the treatment programme being implemented was also identified (Matthey & Črnčec, 2012; Hall et al, 2015).  Out of the remaining papers, four used a RCT design (Hiscock et al, 2008; Price et al, 2012; Hall et al, 2015, Gradisar et al, 2016) and two used a survey rather than an experimental design (Loutzenhiser et al, 2014; Honaker et al, 2018). No explanation was given for the remaining study design (Middlemiss et al, 2012). The choice of a quasi-experimental study design over an RCT is an important consideration as the lack of control group makes it more difficult to ascertain the impact of cause and effect on the independent and dependent variables, and hence are considered less robust sources of evidence (Cluett, 2006).    However, quasi-experimental study designs are often chosen when it is ethically difficult to withhold treatment from a group in need of support (Cluett, 2006), such as not offering sleep interventions to sleep deprived infants and their parents.

Overall all of the studies provided a thorough analysis of the data collection methods used, including an overview of validated and non-validated tools with reasons why these had been chosen and a clear interpretation of the findings.  However, although sample size is also an important ethical consideration only four of the studies provided an analysis of their power calculations and how this had impacted on recruitment to their study (Hiscock et al, 2008; Price et al, 2012; Hall et al, 2015; Gradisar et al, 2016).  Furthermore, acknowledging and managing the researcher impact on the data collection process is important to help reduce the likelihood of researcher bias (Bluff & Cluett 2006); researchers from three of the papers did not have direct contact with participants (Middlemiss et al, 2012; Loutzenhiser et al, 2014; Honaker et al, 2018), blinding was factored into three of the papers (Price et al, 2012; Hall et al, 2015; Gradisar et al, 2016), whereas one paper identified it was impossible to blind the parents or research nurses (Hiscock et al, 2008), two identified that the researchers could have influenced the data collection process and results (Wade et al, 2007; Matthey & Črnčec, 2012) and one did not address the issue (Blunden, 2011).

## Discussion

### Behavioural interventions, infant mental health and maternal mental health

Five out of the ten papers reviewed examined the relationship between behavioural interventions and infant and maternal mental health. Earlier studies tended to focus on infant behavioural cues when assessing infant mental health and the maternal-child bond post intervention and found positive outcomes for infant and maternal mental health (Hiscock et al., 2008; Matthey & Črnčec, 2012). Middlemiss et al.’s (2012) study examined mother and baby physiological parameters and behavioural cues whilst participating in an extinction based inpatient sleep programme, which found that infants’ cortisol levels remained elevated after their behavioural cues had reduced, resulting in a mismatch in the synchrony between the mother and child. Although the mother-child relationship, rather than the sleep programme, were the primary focus of the study, this article led to further debate regarding whether behavioural interventions were harmful (Hiscock & Gradisar, 2012) and is often cited as evidence that these type of strategies increases cortisol levels which may be harmful to the developing brain (AAIMH, 2013; Infant Sleep Information Source, 2018).

Price et al. (2012), followed up Hiscock et al.’s (2008) RCT five years later, using questionnaires, validated tools and salivary samples to analyse whether behavioural interventions, specifically controlled crying, gradual retreat, are associated with harm in the long term.  There were no significant differences in child mental health or chronic stress levels as measured by cortisol, maternal mental health or child-parent bond between the intervention and control groups in this follow up (Price et al, 2012). Furthermore, Gradisar et al.’s (2016) study analysed a range of measures from parents and their infants, including cortisol levels, allocated to either controlled crying group, bedtime fading group or a control group.  This was followed up at 12 months, and parent and child undertook the strange situation test to assess their attachment and no significant differences were found between parents and children in the intervention or control groups (Gradisar et al., 2016).

Overall behavioural interventions recommended in the community, namely controlled crying, gradual retreat and bedtime fading, were found to be effective at treating behavioural insomnias and were not found to be associated with short- or medium-term negative impacts on infant mental health or the parent-child bond and were correlated with positive outcomes for maternal mood (Hiscock et al, 2008; Matthey & Črnčec, 2012; Gradisar et al, 2016). The positive benefits associated with behavioural interventions are estimated to fade two to three years post-intervention, however, no long term negative effects were associated with child mental health or parent-child attachment five years after taking part in the intervention.  One study contradicted these findings (Middlemiss et al, 2012), however, it is important to note that this study used an extinction technique which is rarely recommended in practice and also took place in a residential unit rather than a community setting, which would limit the generalisability of the results.

### Behavioural interventions and maternal mood and/or stress

As half of the mother-child dyad it is important to consider the impact of behavioural interventions on maternal mood, experiences of motherhood and stress.  Mothers of infants with sleep disturbances have a higher incidence of PND compared to the general population, 45-50% versus 10-15%, although it is difficult to ascertain whether mothers experiencing PND are more likely to have infants who develop a sleep issue or infant sleep disorders result in the development of PND (Hiscock & Wake, 2002; Smart & Hiscock, 2007; Črnčec et al, 2009).

Using the Beck Depression Inventory-II, scores for depression reduced in the five participants following the delivery of a group based sleep programme (Wade et al, 2007).  Blunden (2011) adapted a gradual retreat programme to allow parents to respond to their child’s distress and found parental reported levels of satisfaction increased post intervention, including for some parents improved relationship between themselves and their partner and improved mental health for the whole family.  Hall et al.’s (2015) RCT study analysed a range of parental factors in the primary and secondary caregivers such as, depression scores, levels of fatigue and feelings of anger regarding their infant’s sleep, in both the intervention group undertaking controlled crying and the control group who received sleep education. Primary caregivers in the intervention group were found to have reduced depression scores, increased confidence in managing their child’s sleep issues and anger regarding their infant’s sleep issue reduced, compared to the control group (Hall et al, 2015).

### Behavioural interventions and parental perceptions

The majority of research regarding behavioural interventions and infant sleep within the literature is focused on the intervention and the associated effects, rather than the lived experience of the parents undertaking the interventions.  Two of the papers retrieved used internet surveys to question parents about their experiences of implementing behavioural interventions in the community (Loutzenhiser et al, 2014; Honaker et al, 2018). Loutzenhiser et al.’s (2014) study focused on the controlled crying intervention and found a strong association with parental and infant stress when using controlled crying in the community, with over 50% of those using the intervention finding it successful at reducing night wakings.  Those who had a good support network found implementing the technique less stressful, and those who found it more stressful were less likely to report the technique effective (Loutzenhiser et al, 2014). These results differed from Honaker et al.‘s (2018) study which surveyed parents in an online peer support group using one of four techniques; unmodified extinction, extinction, parental presence and parental presence with support, with the majority of parents surveyed reported they had successfully implemented the intervention.  Parental reported levels of stress and infant crying were reported to be high at the beginning of the intervention, but reduced over the first two weeks (Honaker et al, 2018). Both studies found that the majority of parents commence sleep programmes in the community under the current best practice recommended age of six months of age (Loutzenhiser et al, 2014; Honaker et al, 2018). It is important for professionals to be aware that parents may be implementing behavioural interventions in the community earlier than recommended as this can impact on infant distress, maternal mood and infant feeding (Douglas & Hill, 2013).

### Implications for Practice

Due to the contentions in the literature and professional practice, several RCTs have now been conducted to analyse the long-term outcomes of behavioural interventions on infant and maternal mental health and the parent-child attachment and more recently this has also included examining physiological measurements in addition to exploring behavioural cues.  Controlled crying, gradual retreat and bedtime fading have not been associated with levels of chronic infant stress or poor mental health compared to control groups. In addition, maternal mood and levels of parental satisfaction are positively associated with undertaking a behavioural intervention.

An understanding of the lived experience of parents undertaking behavioural interventions in the community is limited, with existing studies highlighting that parents independently implement sleep strategies, starting earlier than recommended, with mixed results.  It is important for professionals supporting families to be aware that they may have already tried to tackle their child’s sleep issues and this may influence their willingness to implement further programmes.

### Limitations of the scope of this review

This rapid review was undertaken to build on an existing review undertaken by the authors to ascertain if new literature had been published to determine the impact of behavioural interventions (Mancz & Wigley, 2017).  Due to time constraints, the screening and study selection was predominately undertaken by one author (GM), although the author erred on the side of caution and included papers that may meet the inclusion criteria and the final selected papers were discussed with the second author (WW). This may have resulted in bias towards the papers selected.

### Conclusions

The existing literature regarding behavioural interventions and the associated outcomes is developing. Several RCTs have not found a link between recommended behavioural interventions, controlled crying, gradual retreat and bedtime fading, and poor infant or maternal mental health or attachment. Further research needs to be undertaken to strengthen the evidence base, particularly when analysing biological markers of stress and attachment.  Additional research is also needed to look into parental perspectives of using behavioural techniques to inform practice and allow practitioners to be mindful of the experiences of the families they are supporting with sleep issues.

The current evidence base indicates that behavioural techniques are not associated with harm to infant mental health or parent-child attachment and can improve maternal mental health, hence, practitioners working with parents of infants with sleep disturbances can confidently recommend these techniques to improve sleeping patterns.

Key points:

* Behavioural insomnias are common in the under-5 age group and are associated with a range of detrimental health outcomes for the child and their wider family.
* As lead professional for this age group, health visitors are ideally positioned to support children and their families with sleep issues, however, there is on-going contention in the literature regarding whether it is appropriate to use behavioural interventions.
* Modified extinction techniques, such as controlled crying and gradual retreat, have not been found to cause long term harm to a child’s mental health or their attachment to their parents and are also associated with an improvement to maternal mental health.

References:

American Academy of Pediatrics (2012) *Infant Sleep Training is Effective and Safe, Study Finds*. Available at: <https://www.aap.org/en-us/about-the-aap/aap-press-room/pages/Infant-Sleep-Training-is-Effective-and-Safe-Study-Finds.aspx>[Accessed 23.8.18]

Australian Association for Infant Mental Health Inc (2013) *Controlled Crying – Position Paper 1*.  Available at: <https://www.aaimhi.org/key-issues/position-statements-and-guidelines/AAIMHI-Position-paper-1-Controlled-crying.pdf>[Accessed 23.8.18]

Bathory, E., & Tomopoulos, S. (2017) Sleep regulation, physiology and development, sleep duration and patterns, and sleep hygiene in infants, toddlers and preschool-age children. *Current Problems in Pediatric and Adolescent Health Care* 47 (2): 29-42

Bayer, J., Hiscock, H., Hampton, A., & Wake, M. (2007) Sleep problems in young infants and maternal mental and physical health. *Journal of Paediatrics and Child Health* 43 (1-2):66-73

Bernard, K., & Dozier, M. (2010) Examining infants cortisol responses to laboratory tasks among children varying in attachment disorganization: stress reactivity or return to baseline.  *Developmental Psychology*. 46 (6): 1771-8

Bluff, R., & Cluett, E. (2006) Critiquing the Literature IN Cluett E and Bluff R (eds) *Principles and Practice of Research in Midwifery*. (2ndEdition). China: Churchill Livingstone – Elsevier 243-261

Blunden, S. (2011) Behavioural treatments to encourage solo sleeping in pre-school children: An alternative to controlled crying. *Journal of Child Health Care* 15 (2): 107-117

Blunden, S., Thompson, KR., & Dawson, D. (2011) Behavioural sleep treatments and night time crying in infants: challenging the status quo. *Sleep Medicine Reviews*. 15 (5): 327-34

Cluett, E. (2006) Experimental research IN Cluett, E. & Bluff, R. (eds) *Principles and Practice of Research in Midwifery*. (2nd Edition). China: Churchill Livingstone – Elsevier 57-89

Črnčec, R., Matthey, S., & Nemeth, D. (2009) Infant sleep problems and emotional health: a review of two behavioural approaches. *Journal of Reproductive and Infant Psychology* 28 (1): 44-54

Douglas, P., & Hill, P. (2013) Behavioural Sleep Interventions in the First Six Months of Life Do Not Improve Outcomes for Mothers or Infants: A Systematic Review. *Journal of Developmental and Behavioural Pediatrics.* 34 (7): 508-522

Gerhardt, S. (2004) why love matters: how affection shapes a baby’s brain. East Sussex: Routledge

Giallo, R., Rose, N., & Vittorino, R. (2011) Fatigue, wellbeing and parenting in mothers of infants and toddlers with sleep problems. *Journal of Reproductive and Infant Psychology* 29(2): 236-249

Gradisar, M., Jackson, K., Spurrier, N., Gibson, J., Whitman, J., Williams, A., Dolby, R., & Kennaway, D. (2016) Behavioural Interventions for Infant Sleep Problems: A Randomized Controlled Trial. *Pediatrics*. 137 (6):e20151486

Gregory, A., & Sadeh, A. (2012) Sleep, emotional and behavioural difficulties in children and adolescents. *Sleep Medicine Reviews* 16 (2):129-136

Haby, M., Chapman, E., Clark, R., Barreto, J., Reveiz, L., & Lavis, J. (2016) What are the best methodologies for rapid reviews of the research evidence for evidence-informed decision making in health policy and practice: a rapid review. *Health Research Policy and Systems* 14: 83

Hall, W., Hutton, E., Brant, R., Collet, J., Gregg, K., Saunders, R., Ipsiroglu, O., Gafni, A., Triolet, K., Tse, L., Bhagat, R., & Wooldridge, J. (2015) A randomized controlled trial of an intervention for infants’ behavioural sleep problems. *BMC Pediatrics*. 15:181

Hill, C. (2011) Practitioner Review: Effective treatment of behavioural insomnia in children. *Journal of Child Psychology and Psychiatry* 52 (7): 731–741

Hiscock, H., Bayer, J., Hampton, A., Ukoumunne, O., & Wake, M. (2008) Long-term Mother and Child Mental Health Effects of a Population-Based Infant Sleep Intervention: Cluster-Randomized, Controlled Trial. *Pediatrics* 122 (3): e621-627

Hiscock, H., & Gradisar, M. (2012) Let’s help parents help themselves: A letter to the editor supporting the safety of behavioural sleep techniques. *Early Human Development* 89 (1): 39-40

Hiscock, H., & Wake, M. (2002) Randomised controlled trial of behavioural infant sleep intervention to improve infant sleep and maternal mood. *BMJ* 324: 1062

Heussler, H., Chan, P., Price, A., Waters, K., Davey, M., & Hiscock, H. (2013) Pharmacological and non-pharmacological management of sleep disturbance in children: An Australian Paediatric Research Network Survey. *Sleep Medicine* 14 (2):189-194

Honaker, S., Schwichtenberg, A., Kreps, T., & Mindell, J. (2018) Real-World Implementation of Infant Behavioural Sleep Interventions: Results of a Parental Survey.  *The Journal of Pediatrics* 199:106-111.e2

Infant Sleep Information Source (2018) *The costs of sleep training*Available at: <https://www.isisonline.org.uk/hcp/how_babies_sleep/sleep_training/cost_of_success/> [Accessed 27.8.18]

Journal of Health Visiting (2018) *About Us*Available at: <http://www.journalofhealthvisiting.com/about.shtml>[Accessed on 24.8.18]

Khangura, S., Konnyu, K., Cushman, R., Grimshaw, J., & Moher, D. (2012) Evidence summaries: the evolution of a rapid review approach. *Systematic Reviews.*  1: 10

Kuhn, B., & Weidinger, D. (2000) Interventions for Infant and Toddler Sleep Disturbance: A Review. *Child & Family Behaviour Therapy* 22 (2): 33-49

Lam, P., Hiscock, H., & Wake, M. (2003) Outcomes of Infant Sleep Problems: A Longitudinal Study of Sleep, Behavior, and Maternal Well-Being. *Pediatrics* 111 (3): e203-207

Loutzenhiser, L., Hoffman, J., &Beatch, J. (2014) Parental perceptions of the effectiveness of graduated extinction in reducing infant night wakings. *Journal of Reproductive and Infant Psychology* 32 (3): 282-291

Mancz, G. & Wigley, W. (2017) Long-term outcomes of techniques used to manage sleep disturbance in the under-5s. *Journal of Health Visiting* 5 (1): 16-24

Martin, J., Hiscock, H., Hardy, P., Davey, B., & Wake, M. (2007) Adverse Associations of Infant and Child Sleep Problems and Parent Health: An Australian Population Study. *Pediatrics* 119 (5): 947-955

Matthey, S., & Črnčec, R. (2012) Comparison of two strategies to improve infant sleep problems, and associated impacts on maternal experience, mood and infant emotional health: A single case replication design study. *Early Human Development* 88(6):437-442

Middlemiss, W., Granger, D., Goldberg, W., & Nathans, L. (2012) Asynchrony of mother-infant hypothalamic-pituitary-adrenal axis activity following extinction of infant crying responses induced during the transition to sleep. *Early Human Development* 88(4):227-232

Mindell, J., Kuhn, B., Lewin, D., Meltzer, L., & Sadeh, A. (2006) Behavioural Treatment of Bedtime Problems and Night Wakings in Infants and Young Children. *Pediatric Sleep* 29 (10): 1263-1276

Mindell, J., & Owens, J. (2009) *A Clinical Guide to Pediatric Sleep: Diagnosis and Management of Sleep Problems* (2ndEdition). China: Lippincott Williams and Wilkins

Mindell, J., Sadeh, A., Wiegand, B., How, T., & Goh, D. (2010) Cross-cultural differences in infant and toddler sleep. *Sleep Medicine* 11 274-280

Moher, D., Liberati, A., Tetzlaff, J., Altman, DG. &, The PRISMA Group. (2009) Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement.  *Annals of Internal Medicine* 151 (4)

Morgenthaler, T., Owens, J., Alessi, C., Boehlecke, B., Brown, T., Coleman, J., Friedman, L., Kapur, V., Lee-Chiong, T., Pancer, J., & Swick, T. (2006) Practice Parameters for Behavioral Treatment of Bedtime Problems and Night Wakings in Infants and Young Children. *Sleep* 29 (10): 1277-1281

Moturi, S., & Avis, K. (2010) Assessment and Treatment of Common Pediatric Sleep Disorders. *Psychiatry* 7 (6): 24-37

Newman, L., Sivaratnam, C., & Komiti, A. (2015) Attachment and early brain development – neuroprotective interventions in infant-caregiver therapy.  *Translational Developmental Psychiatry* 3 (1)

NHS Choices (2012) *Controlled crying ‘safe for babies’*Available at: <https://www.nhs.uk/news/pregnancy-and-child/controlled-crying-safe-for-babies/>[Accessed 23.8.18]

NHS Choices (2016) *Leaving babies to cry ‘will improve their sleep’ study says*Available at: <https://www.nhs.uk/news/pregnancy-and-child/leaving-babies-to-cry-will-improve-their-sleep-study-says/>[Accessed 23.8.18]

Owens, J. (2008) Classification and Epidemiology of Childhood Sleep Disorders. *Primary Care: Clinics in Office Practice*35 (3): 533-546

Price, A., Wake, M., Ukoumunne, O., & Hiscock, H. (2012) Five-Year Follow-up of Harms and Benefits of Behavioural Infant Sleep Intervention: Randomized Trial. *Pediatrics* 130 (4):643-51

Sadeh, A., Mindell, J., & Owens, J. (2011) Why care about the sleep of infants and their parents. *Sleep Medicine Reviews* 15 (5): 335-337

Shore, R. (2003) *Rethinking the Brain: New Insights into Early Development*. New York: Families and Work Institute

Smart, J., & Hiscock, H. (2007) Early infant crying and sleeping problems: A pilot study of impact on parental well-being and parent-endorsed strategies for management. *Journal of Paediatrics and Child Health* 43 (1-2): 284-290

Stevens, A., Garritty, O., Hersi, M., & Moher, D. (2018) *Developing PRISMA-RR, a reporting guideline for rapid reviews of primary studies (Protocol)*.    
Available at: <http://www.equator-network.org/wp-content/uploads/2018/02/PRISMA-RR-protocol.pdf>[Accessed 23.8.18]

Strange, C., Fisher, C., Howat, P., & Wood, L. (2018) ‘Easier to isolate yourself…there’s no need to leave the house’ – a qualitative study on the paradoxes of online communication for parents with young children.  *Computers in Human Behaviour* 83 168-175

Taveras, E., Rifas-Shiman, S., Oken, E., Gunderson, E., & Gillman, M. (2008) Short Sleep Duration in Infancy and Risk of Childhood Overweight. *Archives of Paediatric and Adolescent Medicine* 162(4):305-311

Vriend, J., Davidson, F., Corkum, P., Rusak, B., Chambers, C., & McLaughlin, E. (2013) Manipulating sleep duration alters emotional functioning and cognitive performance in children. *Journal of Pediatric Psychology* 38 (10): 1058-1069

UNICEF (2014) *Building Better Brains: New Frontiers in Early Childhood Development*Available at: <http://www.unicef.org/earlychildhood/files/Building_better_brains____web(1).pdf> [Accessed 22.8.18]

Wade, C., Ortiz, C., & Gorman, B. (2007) Two-Session Group Parent Training for Bedtime Noncompliance in Head Start Preschoolers. *Child & Family Behavior Ther*apy 29 (3): 2007 23-55

Table 1. Overview of the five studies focused on the impact of behavioural interventions on infant and maternal mental health.

Table 2. Summary of the three studies which analysed behavioural interventions and maternal mood and/or stress.

Table 3. Analysis of the two studies exploring parental perceptions of behavioural interventions on their child and themselves.

Figure 1. Flowchart of Study Selection.