NIHR HTA reference 12/150/04 : Development and preliminary evaluation of an intervention package to support parents of excessively crying infants.

[Protocol version 3: for NIHR Project Portfolio].

Project Title: Development and preliminary evaluation of an intervention package to support parents of excessively crying infants.

Research Type: Primary Research

Status: Research in progress

Start date: September 2014

Publication date: January 2017. This is an estimated publication date for this report.

Cost: £443,887.97

Chief Investigator: Professor Ian St James-Roberts

Co-Investigators: Professor Stephen Morris (University College London); Dr Rosemary Garratt (De Montfort University); Professor Susan Dyson (Middlesex University); Ms Nicy Turney (Leicestershire Partnership NHS Trust); Ms Christine Buzzard (Leicestershire Partnership NHS Trust); Dr Elaine Boyle (University of Leicester); Ms Rachel Plachcinski (National Childbirth Trust); Ms Sally Rudge (Consultant Clinical Psychologist); Mr Charles Owen (Institute of Education); Mrs Jan Bullen (Chairperson of Cry-sis).

Contractor: De Montfort University

Plain English Summary

This research is a first step in developing and evaluating new evidence-based NHS services to support parents whose babies cry for prolonged amounts of time. It aims to find out whether a large-scale study is justified.

BACKGROUND: In early infancy, around 1 in 5 babies cry a lot without an apparent reason. This crying used to be known as ‘colic’ and attributed to indigestion pain. Research in the last 20 years has found that only 5-10% of infants taken to the doctor because of prolonged crying are poorly. Most infants who cry a lot are healthy and develop normally. Research has also shown that many normal babies have a crying peak at 1-2 months of age. This and the ‘unsoothable’ crying bouts which alarm parents are linked to normal infant development and stop by 5 months of age.

Although most babies who cry a lot are well, the crying can distress parents and many seek help from their Health Visitor (HV) or GP. The term ‘excessive crying’ refers to this judgement by a parent that the crying is a sign their baby is unwell. It leads to a focus not just on the crying but on parents’ knowledge and vulnerabilities

which affect their ability to provide care, and on subsequent outcomes. Some parents stop breast feeding prematurely because they misinterpret the crying and think their baby is not getting enough to eat. This may hamper infant health, since breast-feeding promotes healthy infant growth and development. The crying can also give rise to maternal depression, poor parent-child relationships, problems with long-term child development, and infant abuse in a small number of cases. Yet, there are no tried and tested NHS practices for supporting parents in managing excessive infant crying. Instead, parents turn to popular books, magazines or websites, which give conflicting advice. By improving services, it should be possible to improve infant outcomes, parents' satisfaction with services, and how NHS money is spent.

THE PROPOSED STUDY has 2 stages. In Stage 1 (Development of an Intervention Package), we will update a 2011 review of the evidence and obtain example materials. For instance, guidelines for supporting parents with crying babies have been developed by an expert international panel. Recently, a nurse-provided group intervention focusing on managing unsettled infants reduced maternal distress, anxiety and depression. Other studies, too, have shown that nurse interventions can help vulnerable parents and improve infant health-care. This first step will generate example components for a support package.

To be successful, the package also needs to be acceptable to parents and to meet their needs. To ensure this, we will run three focus groups, involving 20 parents who have previously reported excessive infant crying. They will be recruited via the study's HV and National Childbirth Trust (NCT) collaborators, asked what they found challenging, what helped, and what should be included in the NHS. Next, with our NHS, NCT, Cry-sis and parent collaborators, we will collate the resulting components into a package suitable for NHS use. We will re-interview the 20 parents and run a workshop with HVs to confirm the suitability of the package. This research stage will decide the form the package will take, but likely components include special training for HVs and leaflets, website/DVD materials and practitioner consultations for parents.

In Stage 2 (Feasibility Study of Package Implementation in the NHS), we will assess whether the new services can be delivered in NHS practice and provide guidance on whether and how a large-scale study of their effectiveness and cost might be carried out. We will recruit two groups. First, parents who approach participant HV clinics for help with infant excessive crying during a 6 month period will be offered the package components and, if written informed consent is given, interviewed to obtain background information and followed up. The two areas of Leicester hosting the study have over 1000 births annually, allowing 30 excessive crying cases to be enrolled during a six month period. However, this number is not certain, while this approach will miss cases where parents judge their baby cries excessively but do not approach HVs, for instance due to lack of knowledge or confidence in themselves or the NHS. Such cases may be especially needy. To overcome these drawbacks, HVs will invite 225 families to enter the study at routine home visits in the first 10 postnatal days. We expect 150 to give informed consent and to be followed up and screened for distress due to excessive infant crying by researchers. This should provide 30 cases for the study. This 'longitudinal' approach may also allow earlier detection and intervention, before problems are entrenched. These two recruitment

strategies should ensure enough participants and indicate numbers and recruitment methods for a future study.

Parents entering the study will be asked to complete short questionnaires to measure parenting stress, anxiety and depression. These will be repeated after the intervention, to indicate potential improvements. We will also ask parents (1) about use of and satisfaction with each package component; (2) suitability of the questionnaires; (3) number and duration of NHS contacts because of infant crying; (4) duration of sole and partial breast-feeding. We will assess how many parents and HVs complete the study. HVs will be asked to rate the package components and barriers/ facilitators to their use. To permit future cost-effectiveness analyses, we will develop measures of costs of each component, together with crying-related NHS costs. A large scale study will be justified if parents and HVs are satisfied with the package, parents remain involved in the study, and the package appears likely to reduce parental distress and be cost-effective in NHS use.

STUDY TIMETABLE: Development of the package components will take 1 year. For the feasibility study, recruitment, intervention and follow-up assessments can be collected in 7 months of year 2, allowing study completion in 24 months.

EXPERTISE IN TEAM: The team includes researchers with experience in studies of infant crying, NHS research, health economic analysis, and Health Visitor/Midwifery training and research. NHS members are Health Visitors and Service Managers in Leicestershire Partnership NHS Trust. Other members are National Childbirth Trust staff, the Chairperson of the charity Cry-sis, parents of previously crying babies, a researcher providing methodological support and Leicester Clinical Trials Unit.

Scientific Summary of Research:

DESIGN: A preliminary study to develop a novel intervention and assess the feasibility of delivering and evaluating it in the NHS.
SETTING: Community Primary Care.
TARGET POPULATION INCLUSION CRITERIA: Parents of any ethnic origin in the Leicestershire Partnership NHS Trust area who speak English, or are supported by English speakers, and have a healthy baby less than 6 months old whose excessive crying causes distress to either parent. English-speaking parents have high distress rates due to infant crying [1] while translation into other languages is not justified at this stage. Local Health Visitor experience is that most non-English-speaking parents are accompanied by English-speaking family members or friends. To prepare for later research, we will record how many non-English speakers are contacted during the study and any obstacles to service delivery that result.
EXCLUSION CRITERIA: Parents of infants who are over 6 months of age or judged ill by Health Visitors or another qualified health professional (these criteria were stipulated in this NIHR HTA call; they make it likely that included infants are healthy).
HEALTH TECHNOLOGY ASSESSED: A support package for parents of infants who cry excessively. The study will have 2 stages:
Stage 1: Development of a Support Package.
The aim at this stage is to develop a novel package of materials that informs and supports parents who judge their baby's crying to be excessive. We will first update a 2011 systematic review of the evidence [2] and obtain a shortlist of example

blueprint materials from research for scrutiny by our participant parents. We cannot complete the shortlist until after that review. However, based on research to date, four components are likely to be included:
(1) A 1-hour briefing workshop for local Health Visitors, since the services are novel and we consider that Health Visitor support and involvement is critical for the success of the study and any future service.
(2) A short information leaflet delivered by the Health Visitors to inform parents about the development of infant crying and its peak in early infancy, the support services available, and our study. The Health Visitors will introduce this leaflet and the services to parents.
(3) More detailed, audio-visual materials about managing infant crying for parents who wish to access them, presented free via website and DVD, with a printed version for parents who prefer it.
(4) A practitioner-delivered group or individual intervention for parents focusing on managing infant crying behaviour, reducing parental distress, anxiety and depression and improving coping.

We have obtained examples of each potential component, together with written permission to use them for our research purposes from the copyright holders. Guidance for supporting parents with crying babies was published by an international expert panel [3] and we have example leaflets from Australian, Canadian/ USA research [4; 5] and from Denmark [6], translated from one which is in routine use in Copenhagen. Digital, audio-visual support materials about infant crying and its management by parents have been developed and evaluated by Australian and Canadian/ USA research [4; 5]. A nurse-provided group brief intervention focusing on managing unsettled infant behaviour reduced maternal distress, anxiety and depression in an Australian study [7]. Strengths included a clear theoretical foundation, involvement of fathers where possible, and evidence of implementation [7]. The use of groups may improve service access and limit costs [8]. Research in related areas, too, shows that nurse or practitioner-provided interventions, particularly based on Cognitive Behaviour Therapy (CBT) rationales, can help vulnerable parents, reduce depression, and may improve infant health-care [9-11]. CBT treatments for acute/moderate adult emotional distress are supported by NICE [12].

Recent reviews highlight parental acceptance and take up of intervention components as crucial for intervention effectiveness [13-14]. To address this, we will run three focus groups, involving 20 parents who have sought NHS help for distress because of excessive infant crying. They will be recruited via the study's Health Visitor collaborators and asked what they found challenging, what helped, about family, community and health service supports, and which components are needed in the NHS. They will be asked to evaluate the shortlisted example materials and advise on their suitability and what modifications are needed. We will collect and analyse the resulting qualitative and rating data. With our NHS, NCT, Cry-sis and parent collaborators, we will collate the resulting components into a package suitable for NHS use. The presentation and educational level of materials will be improved by Institute of Education staff with expertise in development of written and website materials. We will re-interview the 20 parents and run a workshop with Health Visitors to confirm package suitability.

This approach to development of the intervention package is based partly on existing materials with some evidence of suitability and effectiveness, rather than developing every element of every component from the ground up before they are shown to parents. The Canadian/USA website materials, for example, took several years to develop and to repeat this process would be time consuming and expensive. Instead, providing parents with a shortlist of example components to help them to identify the most suitable types of materials is considered to be a more cost-effective strategy. Once we have the parents' feedback, we will develop new materials each evidence-based and with novel content, format and appearance, purpose-designed to be suitable for English parents and the NHS. We will ask Prospect*IP* to confirm that no copyright infringements are involved. Once developed, the leaflet and website materials, in particular, will be inexpensive to deliver. There is evidence that materials of this kind provide parents with information and reassurance that reduce subsequent contacts with health services because of concern about infant crying [15]. If this is confirmed, such materials may be cost-effective in routine health service use and worth evaluating in a future large-scale controlled trial. The direct contact with a trained practitioner will be more expensive to deliver, but likely to be required by a smaller number of parents with greater needs.

Stage 2: Feasibility Study of Package Implementation in the NHS.
We will recruit two groups of parents. First, approximately 80 Health Visitors from four Children's Centres or Sure Start units in Leicester will invite parents who approach them during a 6 month period because of infant excessive crying to participate. Where initial written informed consent is given, they will be interviewed by researchers, offered the package components, and followed up. Estimates are that around 20% of parents judge their baby cries excessively [2; 16]. The Leicester areas targeted for the study have over 1000 births annually, allowing the target 30 cases in a 6 month period.

Although this strategy will allow parents who meet the study criteria to be recruited for the feasibility study, we are concerned not to be entirely dependent on this single recruitment method. One concern is that this method has not been used previously. The estimated prevalence of 20% of cases is based on surveys with unknown recruitment bias carried out several years ago, making it possible that far fewer cases will seek Health Visitor help during a finite period, such as 6 months, and threatening this study. A further concern is that this recruitment method will miss cases where parents do not actively approach Health Visitors for help because of their baby's excessive crying. Here, too, the existing evidence is poor, but there is anecdotal evidence that some parents with an excessively crying baby do not approach medical staff because they feel inadequate and lack confidence in themselves or knowledge of the NHS. Such parents may be especially needy. To overcome these potential threats to the study, Health Visitors will invite 15 families/week to enter the study at the statutory home visit in postnatal days 10-14. We expect 10 per week to give informed consent and to be followed up and screened for distress due to excessive infant crying by researchers, giving a total of 150 families in 15 weeks. This will ensure participants and should identify 30 cases offered the package. This longitudinal approach will also indicate differences between these parents and those who approach Health Visitors for help and may allow earlier detection and intervention, before problems are entrenched. It will also allow us to ask these parents whether they have any reservations about accessing

NHS services for infant crying. These two recruitment methods should indicate potential numbers and recruitment methods for a future RCT, allow adjustment of the recruitment strategy if the expected numbers are not forthcoming, and ensure enough participants. The total of 60 cases will be sufficient for the feasibility study purposes. A chart showing the recruitment and assessment timetable involved in the Feasibility Study on a week-by-week basis is included in Appendix 1.

MEASUREMENT OF OUTCOMES AND COSTS
Parents are likely to be offered the package at around 5 weeks of infant age (the crying peak). We will ask them for demographic data including their socio-economic and educational characteristics and physical and mental health history in the last 2 years, to rate their baby's crying problem severity, and to complete four validated rating scales: the Parenting Stress Inventory; Edinburgh Postnatal Depression Scale; Short Depression, Anxiety & Stress Scale; EQ-5D Quality of Life Questionnaire. We will re-assess parents four weeks after package onset using the same scales, allowing indicative scores and score changes to be considered. Parents will be interviewed: (1) for use of and satisfaction with each package component; (2) suitability of the questionnaires/ suggestions for other measures; (3) number and duration of NHS contacts due to infant crying; (4) duration of sole/ partial breast-feeding and other feeding methods. We will assess how many parents and Health Visitors complete the study. Health Visitors will be asked to keep logs of the lengths and frequency of home visits and other contacts with the study parents and to rate the package components and obstacles/ facilitators to their use.

These procedures will provide measures of the uptake of each package component and parents' and Health Visitors' evaluations of their suitability for use in the NHS, which will indicate whether a future large-scale trial is worthwhile and help to choose the intervention package components and methods for that trial. The questionnaire findings will allow the primary outcome measures to be chosen and decisions to be made about the sample sizes needed to meet statistical power requirements for a full trial. We will ask parents about their willingness to accept randomisation to treatment and control conditions for such a trial. To allow future cost-effectiveness analyses, we will develop measures of costs of each package component (including the Health Visitor and nurse/ practitioner training involved), together with costs of crying-related NHS contact time for existing and new services. A large-scale controlled trial will be justified if parents and Health Visitors are satisfied with the package, take-up is satisfactory, attrition is low, the package appears likely to reduce parental distress and to be effective and cost-effective in NHS use, and parents will be willing to take part.

The arrangements above involve Health Visitors in introducing the study to parents and seeking their provisional consent to take part, while researchers obtain full written informed consent and collect all the measurements, including the summary background data from the Trust electronic records. Two full-time researchers will be needed for this purpose. In addition, the research team will include a qualified CBT practitioner to deliver the intervention needed in the first six months of year 2.

SAMPLE SIZE & PROJECT TIMETABLE (see Flow Diagram): Development of package components will take 1 year. For the Feasibility Study, cases where parents approach Health Visitors for help with excessive crying will be recruited for 6 months,

allowing follow-up to be completed in the first 7 months of year 2. For the longitudinal group, Health Visitors will approach 15 families/week, giving 10 per week (total 150 over 15 weeks) recruited and followed-up in the first 6 months of year 2. A chart showing the Feasibility Study recruitment and assessment timetable is in Appendix 1. This recruitment and assessment schedule is limited by the number of researchers. They will need to enrol or assess up to 20 families per week in the first six months of year 2 (see Appendix 1). However, the expected participant number (N = 60) will be adequate for the purposes of this feasibility study and the design represents a reasonable compromise between the number of parent participants and the cost of the study. The design also allows some flexibility, so that the recruitment methods could be adjusted if numbers prove lower than expected. Data will be entered as collected and data analyses completed and reports generated in a study total length of two years.

TEAM EXPERTISE: The PI has over 20 years experience in conducting successful funded studies of infant crying and its impact on parents and publishing the results in leading peer-reviewed journals, including observational studies, Randomised Controlled Trials and studies in NHS contexts. Other academics in the team have extensive experienced in NHS clinical and health-economic research, clinical trial preparation and management, and in Health Visitor research and training. NHS collaborators are Health Visitors and Service Managers in Leicestershire Partnership NHS Trust, a neonatal paediatrician and active clinical researcher who is joint-lead for the paediatrics speciality group of the Leicestershire Northamptonshire and Rutland Comprehensive Local Research Network, a GP representative, and a clinical psychologist with full qualifications in Cognitive Behaviour Therapy. Other team members are National Childbirth Trust staff, the Chairperson of the charity Cry-sis (which provides telephone counselling for UK parents of crying babies), parents of previously crying babies, and staff from the Thomas Coram Research Unit providing methodological, administrative and Information Technology support. Our overseas collaborators are involved in similar studies in Australia.

Background and Rationale
What is the Problem being addressed?

Surveys in the UK and other countries indicate that around 20% of infants cry for long periods without apparent reason [1-2;16]. This crying used to be attributed to gastro-intestinal disorder and pain, leading to the term ‘colic’ [17]. It is now known that most such infants are healthy and develop normally [18]. Many normal babies have a crying ‘peak’ at 1-2 months of age [1-3]. This and the ‘unsoothable’ crying bouts which alarm parents are linked to normal development and resolve spontaneously by 5 months of age [19].

Although most infants who cry a lot are well, the crying can distress vulnerable parents [20-21] and disrupt their ability to provide care. The term ‘excessive crying’ refers to this judgement by a parent that the crying is a sign their baby is unwell. It has led to a focus not just on the crying but on parents’ responses and subsequent outcomes. For instance, there is evidence that excessive infant crying can contribute to premature cessation of breast feeding because parents misinterpret the crying and think their baby is not getting enough to eat [22]. This may hamper infant health,

since breast feeding promotes healthy infant development [23]. The crying can also trigger depression in vulnerable women, lead to poor parent-child relationships and child development and, in rare cases, precipitate infant abuse [20-21;24-25]. In spite of this background, there are no evidence-based NHS practices for supporting parents in managing infant crying. Instead, parents turn to popular books, magazines or websites, which give conflicting advice. By focusing on how to support parents, health services may reduce parental distress, support parenting and children's development and improve NHS clinical and cost-effectiveness.

The NIHR HTA commissioned call 12/150 reflects this view. Following the call instructions and MRC guidelines [27] we plan a two-stage study. In Stage 1 (Development of an Intervention Package) we will first identify potential components for the package, such as leaflets, website materials, special training for Health Visitors, and brief group interventions, for which evidence of efficacy exists. Recent reviews emphasize that, as well as efficacy, interventions which are effective in practice need to be accepted by parents, so that they act on them [13-14;27]. To this end, we will work with parents who have previously been distressed by their baby's crying to select and improve package components which they consider would help parents in this position and are suitable for NHS use. In Stage 2 (Feasibility Study of Package Implementation in the NHS), we will: (1) assess parents' and Health Visitors' willingness to enter and complete the study; (2) measure parental use and evaluation of the package components; (3) identify outcome measures for assessing package effectiveness; (4) develop cost indices which allow cost-effectiveness analyses; (5) identify barriers/ facilitators to the research; (6) estimate the feasibility and design parameters for a future large-scale study of the intervention package effectiveness and cost.

Evidence explaining why this research is needed now:

Surveys indicate that around 20% of infants cry for long periods without apparent reason [1-2; 16]. This crying used to be attributed to gastro-intestinal disorder and pain, leading to the term 'colic' [17]. It is now known that most such infants are healthy and develop normally [18]. Many normal babies have a crying 'peak' at 1-2 months of age [1-3]. This and the 'unsoothable' crying bouts which alarm parents are linked to normal development and resolve spontaneously by 5 months of age [19].

Although most infants who cry a lot are well, the crying can distress vulnerable parents [20-21] and disrupt their ability to provide care. The term 'excessive crying' refers to this judgement by a parent that the crying is a sign their baby is unwell. This has led to a new focus, not just on the crying but on parents' responses and subsequent outcomes. Excessive infant crying can contribute to premature cessation of breast feeding where parents misinterpret the crying and think their baby is not getting enough to eat [22]. This may hamper infant health, since breastfeeding promotes healthy infant development [23]. The crying can also trigger depression in vulnerable women, lead to poor parent-child relationships and child development and, in rare cases, precipitate infant abuse [20-21;24-25]. Because the crying prompts many parents to seek professional help, the cost to the NHS is substantial [29].In spite of this background, there are no evidence-based NHS services for supporting parents in managing infant crying. By focusing on how to support parents,

evidence-based health services may reduce parental distress, support parenting and children's development and improve NHS clinical and cost-effectiveness.
Drawing on recent research evidence [4-10], this study will develop a package of materials which are likely to support parents in this position. We will then carry out a study to evaluate the feasibility of delivering the package in the NHS, develop methods for measuring its cost-effectiveness, and provide the groundwork for a large-scale, controlled trial of its use.

Aims & Objectives

The aim is to conduct a preliminary study to develop a novel intervention package designed to support parents of excessively crying babies and to examine the feasibility of delivering and evaluating it within the NHS. The research is designed to estimate the feasibility and design parameters for a future large-scale controlled trial of the intervention package's effectiveness and cost-effectiveness.

The study will have 2 stages:
Stage 1: Development of an Intervention Package. Questions to be addressed in this stage are:
1. Based on existing studies and evidence, what are the possible components for a support package for parents of excessively crying babies? For example, possibilities include special training for Health Visitors, leaflets, website/DVD materials and brief group or one-to-one interventions for parents.
2. What types of materials or services do parents of previously crying babies consider would be valuable in providing support, would be accepted by parents, and could be incorporated into the NHS?
3. Is the draft support package developed by this process endorsed by parents and Health Visitors?
Stage 2: Feasibility Study of Package Implementation in the NHS. Questions for this stage of the study are:
1. Is it possible to enrol enough parents of excessively crying babies into a study and retain their participation?
2. What is the optimum recruitment strategy for enrolling parents in the study?
3. Are there differences in the parents recruited by alternative strategies? Might these differences introduce selection bias? What factors enable and inhibit parents of excessively crying babies from contacting the NHS?
4. Is it possible to enrol and retain sufficient numbers of Health Visitors in the study? Do they consider the package worthwhile?
5. Is it feasible for Health Visitors to detect parents who are distressed by their infant's crying?
6. Which package components and outcome measures do parents consider suitable and necessary?
7. Do the outcome measures suggest that exposure to the package components might reduce parental crying-related distress? Which outcome measures perform best?
8. Are parents satisfied with the package and recommend its adoption within the NHS?
9. Is it feasible to measure the costs of each component of the support package and associated NHS costs, including the costs of crying-related contact with Health Visitors and the NHS?

10. Is it feasible to measure the potential benefits of the package in a way which allows cost-effectiveness analysis?
11. What barriers and facilitators are there to the use and evaluation of the package within the NHS?
12. What might be the likely benefits and costs for parents and the NHS of adopting the package?
13. Is it feasible to carry out a large-scale randomised controlled trial, or comparable study, to evaluate the use of the support package in the NHS? What should be the design parameters of such a study? Will parents take part?

Plan of investigation and timetable

Year1 of Project

MONTHS 1-6

- Train Research Officers. Run workshop to familiarise Health Visitors with the study.
- Update systematic literature review and assemble example package components from previous research.
- Develop focus group materials, questionnaires and semi-structured interviews to collect qualitative and rating data from parents.

MONTHS 6-12

- Recruit 20 parents of previously crying babies; obtain written informed consent; carry out 3 focus groups and collect data.
- Collate data and develop preliminary package components, including manual and materials for practitioner-delivered intervention.
- Improve presentation of written or audio-visual (e.g. leaflets, website) materials with help of Institute of Education experts.
- Confirm resulting package components with the parents and Health Visitor collaborators.
- Present interim report to Project Steering Committee.

Year2 of Project

MONTHS 1-7

- Recruit parents of excessively crying infants who approach Health Visitors for help; obtain written informed consent; collect baseline measures, offer and deliver intervention package. Estimated group size n = 30 cases.
- Follow up these Health-Visitor referred cases, collect outcome measures.
- Recruit 150 parents of 10-day-old infants for the longitudinal study group (after allowing for attrition); obtain written informed consent.
- Screen longitudinal group to select parents distressed by excessive infant crying; collect baseline measures; offer and deliver intervention package; follow-up & collect outcome measures. Estimated group size n = 30 cases.

MONTHS 7-12

- Data analysis.
- Present draft final report to Project Steering Committee.
- Run workshops to disseminate findings to NHS professionals and other organisations.
- Submit findings to NIHR, HTA journal and other dissemination outputs.

Design and theoretical/conceptual framework

This is a preliminary study to develop a novel intervention and examine the feasibility of delivering and evaluating it in the NHS, but not formally to evaluate it at this stage. The aims include preparing for, and estimating the parameters of a future controlled trial, but not providing a rehearsal for that trial.

Surveys have found that around 20% of parents in the UK and many other countries are distressed by prolonged and inconsolable infant crying during the first few postnatal months. There are no standard, routine NHS services for helping parents to manage this crying, so that many seek guidance from popular magazines, books or websites. These give conflicting advice, including trying dietary changes and unproven remedies to treat the crying. The term 'infant colic', often used to refer to this crying, represents this traditional view and the assumption that the crying is due to gastro-intestinal disturbance and pain. More recently, studies have found that most such infants are in good health and have normal outcomes, while infants in general have a 'peak' in their crying in the first two months, followed by a pronounced reduction in amount of crying by 3-4 months of age. Although the causes are not yet certain, the evidence points to the conclusion that the crying is linked to normal developmental processes, possibly involving maturational re-organisation of the Central Nervous System, rather than being due to a pathological infant condition.

As well as this change in conceptualisation of the nature and causes of the infant crying, recent studies have highlighted its impact on parents and the poor outcomes which sometimes result. For example, there is evidence that persistent infant crying can contribute to premature cessation of breast feeding because parents misinterpret the crying and think their baby is not getting enough to eat. This may hamper infant health, since breastfeeding promotes healthy infant nutrition. The crying can also trigger depression in vulnerable women, lead to poor parent-child relationships and child development and, in rare cases, precipitate infant abuse.

This recent evidence suggests the need for a different approach to the crying, where the focus is on parental vulnerability and providing support in order to reduce parental distress and improve parent coping, parent-infant interactions and outcomes, rather than on treating a sick infant.

In the longer-term, routine NHS services could include informing and supporting parents who are distressed by their baby's crying in how to manage. Toward that goal, two successive stages of research are needed. The second, and next, stage involves carrying out a Randomised Controlled Trial, or similar large-scale study, to evaluate the new parental support services and assess their clinical effectiveness and cost-effectiveness compared to routine NHS services. That trial will be expensive to carry out and requires preparatory work to develop the package of materials involved in the new service, establish whether it is possible to deliver them in routine NHS conditions, and work out the design and methods to be used in the large-scale trial. Those are the aims of the current study. To ensure that the package developed meets parents' needs, the research at this stage will work closely with parents to choose its components and decide how they are delivered. Once the

package is developed, we will assess which components parents and NHS Health Visitors value and take up in practice and carry out the necessary preparatory work to establish the methods and design for a large-scale trial and to help NIHR decide whether that trial is worthwhile.

An important decision is how far we assess the comparator 'treatment as usual' services at this stage. In principle, we could recruit, assess and follow-up a 'control group' of parents who are distressed by their baby's crying but do not receive the support package, including measuring the amount of NHS time, and associated costs, involved. However, that would require us to recruit fewer parents into the group offered the package or to increase numbers recruited, making this preparatory study more expensive. Since the control group is not central to the development of the support package or the feasibility of its delivery, we have concluded that full direct assessment of a control group of parents will be better left for the more extensive analyses included in a large-scale controlled trial. Instead, the procedures used here will provide the groundwork for that trial by developing methods for assessing and costing the Health Visitor and other NHS services involved. Further, the routine Health Visitor SystmOne electronic records will allow us to collect background data about contacts between parents and Health Visitors where parents report distress because of their baby's excessive crying. We will be able to provide an estimate of the costs involved in such cases, as well as precise figures for the costs involved in delivering the package, using the data available at this stage.

Target population
Parents in the UK of any ethnicity who are distressed by the excessive crying of their infant who is less than 6 months of age. This includes both mothers and fathers where either one of them is distressed by their baby's crying.

Inclusion/exclusion criteria

INCLUSION CRITERIA: Parents of all ethnic communities served by Leicestershire Partnership NHS Trust who speak English, or are supported by English speakers, and have a healthy baby less than 6 months old whose excessive crying causes distress to either or both parents. Local Health Visitor experience is that most non-English-speaking parents are accompanied by English-speaking family members or friends. To prepare for later research, we will record how many non-English speakers are contacted during the study and any obstacles to service delivery that result.
EXCLUSION CRITERIA: infants over 6 months of age or judged ill by Health Visitors or other qualified health professionals.

Setting/context: Community Primary Care.

Sampling

In stage 1, we will recruit 20 parents who have previously sought NHS help for distress because of excessive infant crying to take part in focus groups to help to develop package materials. We will encourage fathers, as well as mothers, to participate. They will be recruited via the study's Health Visitor collaborators and we will ask representatives from Leicester ethnic communities to help to recruit

participants. If needs be, our National Childbirth Trust and Cry-sis collaborators can be asked to recruit parents who meet these criteria.

Stage 2 of the research involves a preliminary evaluation of the parental support package developed in stage 1, including whether it is feasible to deliver and fully evaluate it within the NHS**.** Here, too, the target participants are parents of any ethnicity who have a healthy baby less than 6 months old whose excessive crying causes distress to either or both parents. They will be recruited by approximately 80 Health Visitors from four Children's or Sure Start Centres in Leicester. Two recruitment strategies will be used. First, the Health Visitors will invite parents who approach them during a 6 month period because of infant excessive crying to take part. Where initial written informed consent is given, they will be interviewed by researchers, offered the package components, and followed up. Estimates are that around 20% of parents judge their baby cries excessively. The Leicester areas targeted for the study have over 1000 births annually, allowing the target 30 cases in a 6 month period.

Although this strategy will allow parents who meet the study criteria to be recruited for the feasibility study, we are concerned not to be entirely dependent on this single recruitment method. To overcome this, our participating Health Visitors will also invite 15 families/week to enter the study at the statutory home visit in postnatal days 10-14. We expect 10 per week to give informed consent and to be followed up and screened for distress due to excessive infant crying by researchers, giving a total of 150 families in 15 weeks. This will ensure participants and should identify 30 cases offered the package. This longitudinal approach will also indicate differences between these parents and those who approach Health Visitors for help and may allow earlier detection and intervention, before problems are entrenched. These two recruitment methods should indicate potential numbers and recruitment methods for a future RCT, allow adjustment of the recruitment strategy if the expected numbers are not forthcoming, and ensure enough participants. The total of 60 cases will be sufficient for the feasibility study purposes.

Data collection

The data will be collected by two full-time Research Officers employed for this purpose by the study. During the first year (Stage 1) of the study, they will:

- carry out a literature search to update the 2011 review of interventions to support parents of crying babies;
- help to run the workshop to familiarise the local Health Visitors with the study;
- recruit 20 parents of previously crying babies with the help of the Health Visitors;
- help to assemble the example support package components for consideration by parents;
- develop the materials to carry out the focus groups, help to run the groups and collect the resulting qualitative and rating data;
- help to assemble the components of the support package developed by this process;
- carry out the confirmation of the developed package by parents and Health Visitors;
- help to prepare reports for the project Steering Committee and take part in its meetings.

During year 2 of the study, they will:

- maintain contact with collaborating health visitors and provide them with study materials;
- visit parents who have given consent to be contacted to fully explain the study and obtain full written informed consent;
- maintain contacts with parents, produce copies of assessments and carry out baseline measurements;
- provide parents with the support package components and liaise with the study CBT expert to arrange contacts and meetings;
- complete follow-up assessments and provide all the data for computer entry;
- collect data from Health Visitors, plus background data from the Trust electronic records providing permission is given;
- collect data to allow the cost of services to be measured and cost-effectiveness analyses carried out;
- Help to prepare reports for the project Steering Committee and take part in its meetings;
- Assist with dissemination of the findings and publication of reports.

Data analysis

The first stage of the research will generate qualitative and rating data from parents taking part in focus groups. The data are designed to allow selection of the support package components that the parents consider should be included in the NHS. We will use NVivo software to manage the qualitative data employing thematic analysis [8] to identify and summarise views that are common, or are strongly held by participants from particular ethnic backgrounds. The rating data will take the form of 7-point rating scales (e.g. strongly agree – strongly disagree), where each scale requires judgements about a package component on a dimension, such as clarity of the information, suitability of the presentation, and priority for inclusion in the package. The resulting data are readily summarised using conventional descriptive statistics and the data analysed using parametric or non-parametric statistical methods.

The second stage will provide several types of data:

- summary figures for parental recruitment, take-up of support package components, and persistence in the study;
- summary figures for Health visitor participation and persistence in the study;
- scores from questionnaires and rating scales with established reliability and validity. We will generate descriptive data, to allow the parents' scores for each package component to be summarised and compared with scores from previous studies. In addition, we will use repeated measures statistics to examine score changes from baseline to outcome points.
- frequency and duration data and costs for the package components and existing services, including staff salaries and add-on costs. These will be analysed to assess the amount of staff-parent contact time and associated potential costs for the NHS. Methods will be developed to allow cost-effectiveness analyses. Professor Steve Morris, a co-applicant and expert in health economic analyses, will supervise the data collection and analysis involved.
- summary ratings from parents and Health Visitors to identify the package components and instruments they consider effective and valuable.

It is important to note that this is a preparatory and largely descriptive study, designed to provide estimates for the feasibility and elements of a possible future large-scale controlled trial. The group sizes at this stage are not designed to ensure enough power to allow tests of statistical significance. The purpose of data analysis will be to generate findings that identify promising methods and design parameters for future research. The data coding and analysis will be carried out by Leicester Clinical Trials Unit, which will also advise the study throughout its preparation and conduct. Analysis of the focus group data will be supervised by the Senior Research Officer in the Thomas Coram Research Unit, University of London, who has many years of experience in analysis of qualitative data.

Expected impacts and benefits

Parents with new babies receive statutory visits from Health Visitors at 10-14 days and 6 weeks of infant age, with additional contacts where needed. At these visits, Health Visitors check various health indices and ask about parental concerns in general.

Although prolonged infant crying is a common source of parental concern and distress in the first three postnatal months, there are currently no standard, evidence-based, methods for detecting or assessing these cases or providing parents with information or support. Moreover, Health Visitors and other professionals involved in delivering UK primary health services do not routinely receive any evidence-based instruction in how to support and manage these cases in their foundation or subsequent training courses.

As a consequence of this lack of evidence-based NHS services, parents who are distressed by their baby's crying may seek help from Health Visitors, GPs, hospital Accident & Emergency departments, or other NHS or private sources. Contacts for this reason are common and expensive for the NHS, but these services too are uncoordinated, not evidence based, and highly variable from place to place. Often, too, parents adopt non-prescribed remedies, such as 'colic drops' or herbal remedies, for which there is no evidence of efficacy.

This study will provide the first evidence of the feasibility, reach and uptake of an evidence-based program to support parents of excessively crying babies within the NHS. If the findings prove positive the next stage will be to complete a large-scale study to assess the clinical effectiveness and cost of the new services, compared to the existing NHS provisions. The results at that stage are likely to be of interest to a wide audience, including NHS commissioners, the media, and the general public. The intended long-term beneficiaries are:

- Parents distressed by prolonged unexplained crying in 0-6 month-old babies. The crying is highly aversive to listen to. The figures depend on the definitions used, but surveys in the UK and other countries indicate that this involves approximately 20% of parents to some degree. Many parents cope, but lack of supports can result in feelings of helplessness, guilt where parents feel they are inadequate parents, depression in vulnerable women and frustration and exhaustion in others.

- The infants themselves, since the crying can lead parents to stop breast-feeding prematurely because they think their baby is hungry. This then hampers healthy infant growth and conflicts with NHS policy for infant nutrition. In some cases, the parents' exhaustion or depression leads to inadequate parent-child interaction which persists after the crying stops, with adverse consequences for the infant's development. In extreme cases, the crying can precipitate infant abuse.
- The Health Visitor profession. In the UK, Health Visitors provide the primary source of NHS support for parents and babies. Development of the profession has become a government priority, but as well as more Health Visitors their work needs to be evidence-based. This is not currently the case in relation to providing parents with information and support about prolonged infant crying and how to manage it. The Institute of Health Visiting, recently set up to promote excellence in Health Visiting, is a collaborator in this study.
- The NHS, since the findings from this research are designed to improve the clinical effectiveness and cost-effectiveness of NH Services for families. Because around 20% of UK parents are distressed by excessive infant crying and the NHS costs involved are estimated to be over £65 million per year, the new services have the potential to help many families and increase NHS clinical and cost-effectiveness to a substantial degree.

Dissemination

The main methods of dissemination planned at this stage are:

- We will run two workshops for Primary Health Professionals to report the findings, one in London and one in Leicester, both open to other professionals and areas of the UK;
- The Leicestershire Northamptonshire and Rutland Comprehensive Local Research Network will distribute notification of the study and assist in disseminating findings;
- The Institute for Health Visiting will include an announcement about the study, and a report of the findings, in its newsletter, as well as disseminating via its twitter page and links with *netmums* and the Royal Society of Public Health;
- The National Childbirth Trust will disseminate information about the study and its findings via its websites, professional network, intranet, press office and conferences;
- The study and findings will be listed in Chimat Knowledge Update and other internet sites and networks;
- We will present the findings at national and international conferences;
- We will submit a report of the findings to the NIHR HTA;
- We will submit articles for publication to the HTA journal and to professional journals read by Health Visitors and other Primary Health Professionals.

Project management

We will establish an independent Study Steering Committee following the NIHR Research Governance Guidelines to monitor the project.

The Study Management Group (SMG) will meet 2-3 times/year to receive reports from the research team, provide support, and monitor progress. Members and areas of responsibility are as follows:

- The Senior Research Officer employed by the project will be responsible for the day-to-day running of the project and for the Junior Research Officer who will assist her/him.
- Rosemary Garratt, Principal Lecturer in the Midwifery Research Centre, De Montfort University, will manage the project and supervise the Research Officers.
- Sue Dyson, until recently Director of Nursing & Midwifery Research at De Montfort University, will supervise ethical procedures and provide overall guidance. Dr Dyson became Professor of Nursing, Middlesex University in May 2013.
- Elaine Boyle is a paediatrician and Senior Lecturer, Department of Health Sciences, University of Leicester. Dr Boyle is also joint-lead for the paediatrics speciality group of the Leicestershire Northamptonshire and Rutland Comprehensive local Research Network. She will oversee infant health issues.
- Sally Rudge is a consultant clinical psychologist with CBT qualifications employed by Northamptonshire Healthcare NHS Trust. She will advise on the intervention package, including the practitioner-delivered intervention included in year 2 of the study.
- Nicy Turney, Senior Nurse-Professional Lead Health Visiting in the Families and Young Peoples Division of Leicestershire Partnership NHS Trust, will supervise the Health Visitors involved in the project.
- Chris Buzzard, Service Manager, Health Visiting and School Nursing, in the Families and Young Peoples Division of Leicestershire Partnership NHS Trust, will co-supervise the Health Visitors involved in the project.
- Raksha Pandya-Wood, Senior Research Fellow and Regional Lead in User Involvement in the East Midlands NIHR RDS will provide guidance on recruitment of ethnic groups into the study.
- Rachel Plachcinski, Senior Academic Liaison Officer at the National Childbirth Trust (NCT) will represent the NCT on the SC and liaise with NCT volunteers in the Leicester area.
- Stephen Morris, Professor of Health Economics in the Research Department of Epidemiology & Public Health, University College London, will be responsible for the design and supervision of the health economics parts of the study.
- Leicester Clinical Trials Unit will provide guidance on trial management and process and analyse the study's quantitative data.
- Ian St James-Roberts, Professor of Child Psychology in the Institute of Education, University of London, is the study PI. He will oversee the study as a whole.
- Charlie Owen is Senior Research Officer, Thomas Coram Research Unit, Institute of Education, University of London. He will oversee the qualitative data analysis and chair the SMG.
- Dr Zahida Adam, MA (Oxon), BMBCh, MRCGP, DRCOG will represent GPs in the Leicester area.
- Two parents who have previously had infants who cried excessively will be invited on to the SMG.
- Representatives from Leicester ethnic communities and Health Visitors will be invited onto the SMG.

Rosemary Garratt and Ian St James-Roberts will be responsible for maintaining communication between members of the project team.

Approval by ethics committees.

We are seeking ethical approval from De Montfort University Ethics Committee, together with medical research ethics approval through the IRAS system. Co-applicant Sue Dyson, a previous member of Leicestershire, Northamptonshire and Rutland Ethics Committee 2, will guide our submission through the ethical review process.

Patient and public involvement

Parents who have previously been distressed by their infant's crying will be participants in the study and will be invited to be members of the Study Management Group. In the first stage, they will help in choosing the components for a support package to support parents of excessively crying infants. In stage 2, other parents will be invited to take part in the feasibility study designed to evaluate the package components in routine use. Their take-up and ratings of the package components will provide key data. Representatives of ethnic and cultural communities in Leicester will be informed about the study and included in the SMG.

Expertise and justification of support

The study PI, Ian St James-Roberts is Professor of Child Psychology in the Institute of Education, University of London, with extensive experience carrying out successful funded research studies of infant crying and its impact on parents and reporting the results in leading peer-reviewed journals. This includes Randomised Controlled Trials, comparative studies and research in NHS contexts. He has written a textbook on evidence-based services for infant crying and sleeping problems for Primary Health Professionals [1].
Stephen Morris is Professor of Health Economics in the Research Department of Epidemiology & Public Health, University College London. He has extensive experience in health-economic studies in the NHS. He will supervise the design and conduct of the study health economic analyses.
Rosemary Garratt is Principal Lecturer in the Midwifery Research Centre, De Montfort University, with expertise in Midwifery training, postgraduate research supervision and project management. She will manage the study and Research Officers.
Sue Dyson, until recently Director of Nursing & Midwifery Research at De Montfort University, will supervise ethical procedures and provide overall guidance. She has been involved in the study from its inception and has extensive NHS, academic, ethics and community networks in Leicester. She became Professor in Nursing, Middlesex University in May 2013.
Nicy Turney, Senior Nurse-Professional Lead Health Visiting in the Families and Young Peoples Division of Leicestershire Partnership NHS Trust, will supervise the Health Visitors involved in the project.
Chris Buzzard, Service Manager, Health Visiting and School Nursing, in the Families and Young Peoples Division of Leicestershire Partnership NHS Trust, will co-supervise the Health Visitors involved in the project.

Charlie Owen is the Senior Research Officer, Thomas Coram Research Unit, University of London. He has extensive experience of research methodology and qualitative data analysis.
Rachel Plachcinski is Senior Academic Liaison Officer at the National Childbirth Trust (NCT) and a qualified practitioner. She will represent the NCT and liaise with its Leicester branch.
Dr Elaine Boyle, a paediatrician and Senior Lecturer in Neonatal Medicine, Department of Health Sciences, University of Leicester, will oversee paediatric issues. Dr Boyle is also joint-lead for the paediatrics speciality group of the Leicestershire Northamptonshire and Rutland Comprehensive Local Research Network.
Sally Rudge is a consultant clinical psychologist with CBT qualifications employed by Northamptonshire Healthcare NHS Trust. She will oversee the development of the intervention package, including the practitioner-delivered intervention we expect to include in the study.
Jenny Hogg, Communications & Marketing Coordinator in the Institute of Education has extensive experience in preparing and ensuring the educational suitability of online and other media materials. She will assist in the development of the support package and ensure the suitability of its materials.
Jan Bullen is Chairperson of the national charity Cry-sis, which provides telephone support for parents with crying babies. She will advise the project and scrutinise the intervention package it develops.
Leicester Clinical Trials Unit will advise on the study methods and data collection, store and analyse the quantitative data, and ensure that suitable evidence is collected to prepare the way for a possible future large-scale controlled trial.
Dr Zahida Adam, MA (Oxon), BMBCh, MRCGP, DRCOG will represent GPs in the Leicester area.
Our overseas collaborators are:
Dr Harriet Hiscock, Principal Fellow in the Department of Paediatrics, University of Melbourne, who has carried out extensive research into health services for infant crying and sleeping problems.
Professor Jane Fisher and Dr Heather Rowe, School of Public Health & Preventive Medicine, Monash University. They have published provisional findings from research developing support materials for parents of excessively crying babies [7]. Their collaboration includes providing example materials for the support package and advising throughout the research.

References

1. St James-Roberts I (2012). The Origins, Prevention and Treatment of Infant Crying and Sleeping Problems: an evidence-based guide for healthcare professionals and the families they support. Routledge, London.
2. Douglas P, Hill P (2011). Managing infants who cry excessively in the first few months of life. BMJ**,** 343, 1265-1269 doi: 10.1136/bmj.d7772.
3. Barr RG, St James-Roberts I, Keefe M (Eds. 2001). New Evidence on Unexplained Early Infant Crying: Its Origins, Nature and Management. Johnson & Johnson Pediatric Institute Round Table, Skillman, NJ.
4. Barr RG, Barr M, Fujiwara T, Conway J,Catherine N, Brant R (2009).Do educational materials change knowledge and behaviour about crying and shaken baby syndrome? A randomized controlled trial. Canadian Medical Association Journal; 180. DOI:10.1503/cmaj.081419.
5. Cook F, Bayer J, Le HND, Mensah F, Cann W, Hiscock H. Baby Business: a randomised controlled trial of a universal parenting program that aims to prevent early infant sleep and cry problems and associated parental depression. BMC Pediatrics 12:13. DOI:10.1186/1471-2431-12-13
6. Alvarez M. & Talbro A (2010). Hvorfor graeder barnet? (Why is this baby crying?). Komiteen for Sundhedsoplysning, Kopenhavn, Denmark.
7. Fisher JRW, Wynter KH, Rowe HJ (2010). Innovative psycho-educational program to prevent common postpartum mental disorders in primiparous women: a before and after controlled study. BMC Public Health, 10:432. http://www.biomedcentral.com/1471-2458/10/432.
8. Stevenson MD, Scope A, Sutcliff PA, Booth A, Slade P, Saxon D, Kalthenthaler E (2010). Group cognitive behavioural therapy for postnatal depression: a systematic review of clinical effectiveness, cost-effectiveness and value of information analysis. Health Technology Assessment, 14 (44), 1-107, iii-iv, doi 10.3310/hta14440.
9. Barnes J, Ball M, Meadows P, Howden B, Jackson A, Henderson J, Niven L (2011). The Family-Nurse Partnership programme in England. Wave 1 implementation in toddlerhood and a comparison between waves 1 and 2a of implementation in pregnancy and infancy. London: Department of Health.
10. Renfrew MJ, McCormick FM, Wade A, Quinn B, Dowswell T (2012). Support for healthy breastfeeding mothers with healthy term babies. Cochrane Database of Systematic Reviews 2012, Issue 5. Art. No.: CD001141. DOI: 10.1002/14651858.CD001141.pub4.
11. Kemp L, Anderson T, Travaglia J, Harris E (2005). Sustained nurse home visiting in early childhood: exploring Australian nursing competencies. Public Health Nursing, 22: 254–259. DOI: 10.1111/j.0737-1209.2005.220309.x
12. NICE Clinical Guidelines CG90 (2009). Depression in Adults: The Treatment and Management of Depression in Adults. www.nice.org.uk
13. Moore TG, McDonald M, Sanjeevan S, Price A (2012). Sustained home visiting for vulnerable families and children: A literature review of effective processes and strategies**.** Parkville Victoria: The Royal Children's Hospital's Centre for Community Child Health and the Murdoch Children's Research Institute.
14. Olds DL, Sadler L, Kitzman H (2007). Programs for parents of Infants and toddlers: recent evidence from randomised trials. Journal of Child Psychology & Psychiatry, 48, 355-391.
15. St James-Roberts I, Sleep J, Morris S, Owen C, Gillham P (2001). Use of a behavioural programme in the first three months to prevent infant crying and sleeping problems. Journal of Paediatrics & Child health, 37, 289-297.
16. St James-Roberts I, Halil T (1991). Infant crying patterns in the first year: normal community and clinical findings. Journal of Child Psychology & Psychiatry, 32, 951-968.
17. Illingworth RS (1985). Infant colic revisited. Archives of Disease in Childhood, 60, 981-985.
18. Lehtonen L (2001). From colic to toddlerhood. In Barr RG, St James-Roberts I, Keefe M (Eds.). New Evidence on Unexplained Early Infant Crying: Its Origins, Nature and Management. Johnson & Johnson Pediatric Institute Round Table, Skillman, NJ. Pp 259-272.
19. Barr RG, Paterson J, MacMartin L, Lehtonen I, Young S (2005). Prolonged and unsoothable crying bouts in infants with and without colic. Developmental & Behavioral Pediatrics, 26, 14-22.
20. Murray L, Cooper P (2001). The impact of irritable infant behavior on maternal mental state: a longitudinal study and treatment trial. In Barr RG, St James-Roberts I, Keefe M (Eds.). New Evidence on

Unexplained Early Infant Crying: Its Origins, Nature and Management. Johnson & Johnson Pediatric Institute Round Table, Skillman, NJ. Pp 149-164.

21. Vik T, Grote V, Escribano J, Socha J, Verduci E, Fritsch M, Carlier C, von Kries R, Koletzko B (2009). Infantile colic, prolonged crying and postnatal depression. Acta Paediatrica, 98, 1344-1348
22. Howard CR, Lanphear N, Lanphear BP, Eberly S, Lawrence RA (2006). Parental responses to infant crying and colic: the effect on breastfeeding duration. Breastfeeding Medicine, 1(3):146-55.
23. Beake S, Pellowe C, Dykes F, Schmied V, Bick D (2012). A systematic review of structured compared with non-structured breastfeeding programmes to support the initiation and duration of exclusive and any breastfeeding in acute and primary health care settings. Maternal and Child Nutrition, 8, 141–161.
24. Papousek M, von Hofacker N (1998). Persistent crying in early infancy: a non-trivial condition of risk for the developing mother-infant relationship. Child: Care, Health & Development 24, 395-424.
25. Lee C, Barr RG, Catherine N, Wicks A (2007). Age-related incidence of publicly reported shaken baby syndrome cases: is crying a trigger for shaking? Journal of Developmental & Behavioral Pediatrics, *28*, 288–293.
26. Kitzinger J. (1995) 'Introducing focus groups', *British Medical Journal* 311: 299-302.
27. MRC Guidelines for Developing and Evaluating Complex Interventions: New Guidance (2007). http://www.mrc.ac.uk/Utilities/Documentrecord/index.htm?d=MRC004871.
28. St James-Roberts I (2006) Infant crying and sleeping problems: recent evidence for best practice. Workshop for UK Primary Healthcare Service Professionals, De Montfort University Leicester, May 12; repeated for professionals in the United Bristol NHS Trust, Bristol, October 4 2007.
29. Morris S, St James-Roberts I, Sleep J, Gillham P (2001). Economic evaluation of strategies for managing crying and sleeping problems. *Archives of Disease in Childhood*, 84, 15–19.

APPENDIX 1: Recruitment and Assessment Chart for Stage 2 of the Research (The Feasibility Study. Please see KEY below)

Study Stage 2: Weeks in Year 2	Excessive infant crying cases recruited where parents approach Health Visitors (HV arm)	Longitudinal arm cases Recruited @ 10 days infant age	Excessive crying cases selected from longitudinal arm @ 5 weeks age (20% prevalence)	Parents offered package		Outcome measures 4 weeks later	
				HV cases	Long'l cases	HV cases	Long'l cases
1	1	10		1			
2	2	10		2			
3	1	10		1			
4	2	10		2			
5	1	10	2	1	2	1	
6	2	10	2	2	2	2	
7	1	10	2	1	2	1	
8	2	10	2	2	2	2	
9	1	10	2	1	2	1	2
10	2	10	2	2	2	2	2
11	1	10	2	1	2	1	2
12	2	10	2	2	2	2	2
13	1	10	2	1	2	1	2
14	2	10	2	2	2	2	2
15	1	10	2	1	2	1	2
16	2	N = 150	2	2	2	2	2
17	1		2	1	2	1	2
18	2		2	2	2	2	2
19	1		2	1	2	1	2
20	2		N = 30	2	N = 30	2	2
21	1			1		1	2
22	2			2		2	2
23	1			1		1	2
24	2			2		2	N =30
25	N = 36			N ≥ 30		1	
26						2	
27						1	
28						N ≥ 30	

KEY: We expect 1 or 2 Health Visitor (HV) cases to be recruited per week (please see the main text for particulars). They will be offered the package then followed up 4 weeks later, allowing follow-up assessment of this group in 28 weeks. Cases for the longitudinal arm will be recruited at 10-14 days infant age. We will recruit 150 longitudinal cases (after attrition). In 20% of these cases, we expect at least one parent to be distressed by infant crying at around 5 weeks of infant age (the infant crying peak). They will be offered the package then followed up 4 weeks later, allowing outcome assessments of these cases in approximately 23 weeks.