Infants that cry excessively: The effect of regularity and swaddling

Bregje Elisabeth van Sleuwen

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Wensvlaggen of gebedsvlaggen worden opgehangen om de lucht te zuiveren en goden gunstig te stemmen. Wanneer de vlaggen wapperen in de wind worden de wensen of gebeden meegenomen naar de hemel. De kleuren hebben ieder een eigen betekenis. Rood – vuur, groen – hout, geel – aarde, blauw – water, wit – ijzer. Het Tibetaanse woord voor wensvlaggen is Lung Ta.

Lay-out: Bregje Elisabeth Raap – van Sleuwen.
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Stellingen behorende bij het proefschrift

Infants that cry excessively
The effect of regularity and swaddling

Utrecht, 28 mei 2008

Bregje Elisabeth van Sleuwen

1. Inbakeren werd in toenemende mate toegepast in Nederland zonder dat bekend was of dit van invloed is op het huilen van baby's. Dit onderzoek toont aan dat inbakeren geen toegevoegde waarde heeft naast het bieden van regelmaat, voorspelbaarheid en prikkelreductie. (*Dit proefschrift*).

2. Het toepassen van regelmaat, voorspelbaarheid en prikkelreductie, al dan niet aangevuld met inbakeren, kost weinig tijd, de begeleiding wordt zeer gewaardeerd door ouders en de adviezen zijn eenvoudig op te volgen. (*Dit proefschrift*).

3. Een afname van huilen met 42% na één interventieweek is voor ouders van excessief huilende zuigelingen en hun hulpverleners van zeer grote waarde. (*Dit proefschrift*).

4. Inbakeren kan veilig toegepast worden wanneer het vergezeld gaat van advies over slaappositie, de techniek waarmee het kind wordt ingebakerd (hoofd dient vrij te blijven), de hoeveelheid beddengoed en hoe strak de doeken rondom de borst, knieën en heupen worden aangebracht. (*Dit proefschrift*).

5. Bij vermoeden van een koemelkeiwit allergie hebben ouders meer steun nodig bij het doorlopen van een eliminatie provocatie-re-eliminatie test met hun kind. Zolang deze test niet (volledig) uitgevoerd wordt, blijft het onduidelijk hoe groot het percentage kinderen is dat daadwerkelijk een koemelkeiwit allergie heeft. (*Dit proefschrift*).

6. Het feit dat er zo weinig psychologische verschillen waarneembaar zijn tussen moeders van excessief huilende zuigelingen en moeders van excessief huilende zuigelingen die positief reageerden op een hypoallergeen dieet suggereert dat het hoge aandeel van beide groepen moeders in het klinisch gebied op psychologische meetinstrumenten in vergelijking met controlemoeders een gevolg is van het excessieve huilen en niet de oorzaak van het overmatige huilgedrag. (*Dit proefschrift*).

7. Het feit dat beide interventies geen verschil laten zien in effect tussen moeders van excessief en matig huilende zuigelingen, suggereert dat beide interventies toepasbaar zijn voor alle moeders wiens zuigeling overmatig huilt, onafhankelijk van de hoeveelheid huilen. (*Dit proefschrift*).

8. Excessive crying can be seen as an objectively measurable phenomenon, which either does or does not fulfil the criteria of Wessel, or as a parental complaint, in which parents decide that the amount of crying is abnormal.
Lehtonen LA, Rautava PT. Curr Probl Pediatr 1996;26:79-85

9. The intense crying and inconsolability of an excessively crying or fussy infant creates a host of parental reactions and concerns about the behavioural development of their child.
Stifter CA. Encyclopedia on Early Childhood Development [online] 2005:1-7

10. Treatment of persistent colic symptoms in infants who are referred to secondary care should be primarily targeted to at support for parents, and not at an underlying medical condition (including cow's milk intolerance) in the infant.
Zwart P, Vellema-Goud MGA, Brand PLB. Acta Pædiatrica 2007;96:401–5

11. Crying is the initiating stimulus reported in many cases of infant abuse and infanticide.
Frodi A. In: Lester BM & Boukydis CFZ (Eds.). New York, 1985

12. Absence of evidence is not evidence of absence. We need to report uncertain results and do it clearly.
Alderson Ph. BMJ 2004;328:476-7

13. If you obey all the rules, you miss all the fun.
Katharine Hepburn, US actress

14. If you educate a woman, you educate a family.
Paul Rosenmöller, oud-leider Groen Links, Volkskrant 2 september 2003

15. De meest bekende potentiële risicofactor voor excessief huilen is het snijden van een ui.
Dr. A.C. Engelberts, kinderarts, 2006

16. Nederlanders mogen best wat meer baby's maken.
Mr. A. Rouvoet, minister voor Jeugd en Gezin, De Pers 19 februari 2008

Infants that cry excessively
The effect of regularity and swaddling

Baby's die excessief huilen
Het effect van regelmaat en inbakeren
(met een samenvatting in het Nederlands)

Proefschrift

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Bregje Elisabeth van Sleuwen

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te Heeswijk – Dinther

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Aan mijn ouders.
Voor Wilbert en onze meiden, Juul en Guusje.

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List of abbreviations

SIDS	Sudden Infant Death Syndrome
DDH	Developmental Dysplasia of the Hip
RCT	Randomised, Controlled Trial
HR	Heart Rate
QS	Quiet Sleep
REM	Rapid Eye Movement
SpO2	Pulse Oxygen Saturation
VLBW	Very Low Birth Weight
R	Regularity
RS	Regularity and Swaddling

General introduction

1

Bregje E. van Sleuwen

Normal crying

Crying is one of the earliest and most powerful, sometimes the only means available to the infant for communicating needs to a caregiver. This "acoustical umbilical cord", as it has been called[1], serves to connect the infant to a caregiver, its source of nurture and protection. The infant itself is helpless to meet his own needs, and thus crying has been posited as an adaptive strategy in the species' struggle for survival over the course of human evolution.[2]

During the first months of life, predictable changes in the amount and the pattern of crying can be seen, despite considerable individual variability. This is called "the early crying curve" (Figure 1). From birth on, the amount of crying gradually increases until it peaks at six weeks, then decreases until 12 weeks, at which time it remains fairly stable until the end of the first year.[3-6] Furthermore, a diurnal pattern, which shows a concentration of crying in the evening hours, is distinguishable as early as ten days of life.[7] This basic appearance of the early crying curve seems to be unaffected by changes in care taking style, in contrast with the amount of crying later in the first year and onward.[7] Therefore, early crying is thought to reflect physiological and maturational transitions during the first months of life. The reduction of crying after six weeks of age coincides with an increase in emotion control of the infant, for example the start of responsive or social smiling.[8]

In Western countries, the average cry/fuss duration during the crying peak at six weeks is 2-2 ½ hours per day.[7] After 3 months of age, infants cry on average about 1-1 ½ hour a day.[9]

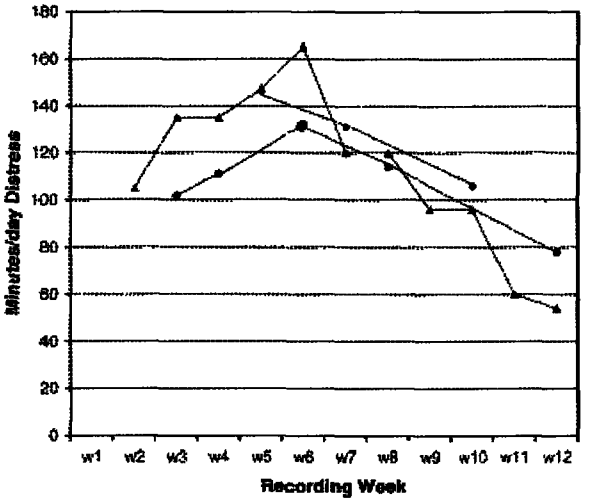

Figure 1. Crying amounts and patterns from three North American studies illustrating absence of secular trend. Triangles: data from Brazelton (1962)[3]; circles: data from Hunziker and Barr (1986)[6]; diamonds: data from Kramer et al. (2001).

Reprinted from Barr RG, Trent RB, Cross J. Age-related incidence curve of hospitalized SBS cases: convergent evidence for crying as a trigger to shaking. Child Abuse Negl 2006;30:7-13.

Excessive crying

Definition

The most widely cited criteria of excessive crying were described by Wessel and his colleagues in 1954 on the basis of a careful, systematic study of infants from The Yale Rooming-In Project.[10] Wessel's commonly known 'rule of three's' defined a "fussy" infant as "one who, otherwise healthy and well-fed, had paroxysms of irritability, fussing or crying lasting for a total of three hours a day and occurring on more than three days in any one week". An infant was seriously "fussy" if, in addition, "their paroxysms continued to recur for more than three weeks, or became so severe that the paediatrician felt that medication was indicated". In 1995, this definition was broadened to the 'modified Wessel's criteria' (crying for three or more than three hours a day for more than three days a week).[11] Excessive crying can be seen as an objectively measurable phenomenon, which either does or does not fulfil the criteria of Wessel, or as a parental complaint, in which parents decide that the amount of crying is abnormal.[12] The group of infants that cry a lot according to their parents can be divided into a group of moderately crying infants (less than 3hrs/24hr) and a group of excessively crying infants (crying for 3 hours or more / 24hrs).[13-16] To meet the need to measure the amount of crying, Barr developed and validated a 'Parental diary of cry and fuss behaviour'.[17] The perception of crying differs from person to person, as well the stress it causes. To measure these subjective feelings, Lester developed the 'Cry Perception Scale'.[18]

Prevalence

The prevalence estimates of excessive crying depend on what definition is used.[19,20] Crying according to the definition of Wessel occurred in 2-2.5% of the Dutch infant population.[9,21,22] In two large Dutch prevalence studies, 13% and 8.6% of parents reported that their infant cried for more than 3 hours for at least one day in the preceding week.[9,21] Of all parents who reported that their infants cried more than 3 hours a day, 32 – 39% actually cried this amount according to the baby day diary.[21,23] Community studies showed a prevalence of maternal distress/concern about their 1-3 month old infants' crying of 12% in Sweden[24], 14% in the Netherlands[20], 19% in Denmark[25], 20% in England[26], and 23% in the USA[27,28].
In the Netherlands, there are 15 scheduled visits (13 at the well-baby clinic and two home visits) within the first 4 years of a child's life. The children are alternatively seen by a youth health doctor and –nurse. Between the age of 4 and 19 years, children visit a school doctor or –nurse two to four times. During these visits, the infants' development and general health are checked and followed. Youth health professionals are easily accessible for information and advice. In the Netherlands, 22% of the parents consulted a youth health professional for their excessively crying infant, in 5% of the cases they consulted a general practitioner, 2% of these infants was presented to a paediatrician, and 6% received other support.[9] Prevalence of

parental approach to health services due to crying was 5% in Finland[29], 15% in Denmark[25], and 15 to 21% in England[26.28,30]

Causes

Various causes are described in the literature for excessive crying, such as cow's milk allergy, gastro-oesophageal reflux, infections, or various rare conditions, but in most cases excessive crying remains unexplained.[31,32] Possible explanations of early excessive crying can be divided into three categories: 1. organic diseases, 2. parental incompetence, and 3. infant temperament/development, as elaborated by St. James-Roberts.[28,33]

"Ad.1. In 1 – 3 month old babies, there is convincing evidence cow's milk protein intolerance can cause crying, although it is rare: it occurs in 1-2% of all infants and in 10% of excessively crying infants.[34,35] Parents themselves often have the impression that excessive crying is caused by intrinsic factors, such as an allergy or physical abnormality".[36]

Ad. 2. As early as in 1957, Breslow hypothesised that "marked evidence of emotional instability on the part of the parents" may be causal in the development of infant crying, particularly for those infants who did not react to formula changes.[37] Furthermore, as St. James-Roberts describes, "challenges to the explanation about parental incompetence are that home observations have shown parents of 'crying babies' to be highly responsive and sensitive.[38,39] Furthermore, home observations and laboratory studies have shown that the babies are objectively hard to soothe.[40,41] By rapid response or constant intervention, parents can reduce the crying and translate it into fussing, but the problem of unsoothable crying will not disappear.[41]

Ad. 3. Explanations for this assumption are the neuro-behavioural shift that occurs in the first 3 months of age, which involves a maturation-driven change in central nervous system (CNS) controlling behaviour. Furthermore, the explanatory framework is re-located: from gut to CNS; from pathology to normal growth and development. But if crying is normal in development, why do many infants cry relatively little, and others a great deal more? Obviously, there must be individual differences in neuro-development".[33]

Predictors

White et al. suggested in a case-control study a causative contribution of dysregulation of the hypothalamic-pituitary-adrenal (HPA) axis: the normal circadian rhythm in cortisol production was absent in excessively crying infants (those that cry for more than 3 hours a day).[40] An association of dysregulation of the HPA axis can also be found in other emotional and behavioural problems in children, such as depression[42] and ADHD[43,44]. Antenatal stress, anxiety, and depression are hypothesized to be of influence on postnatal changes in HPA axis activity and associated development disturbances. Due to these factors, maternal glucocorticoid hormones, such as cortisol, are enhanced and might (permanently) affect the fetal

neuroendocrine systems, especially the HPA axis.[45] As a consequence, due to the altered functioning of the fetal HPA axis, long term effects in brain morphology, physiology, and behaviour (fetal programming) might be seen.[46]
A large prospective Dutch community-based study recently demonstrated that women who have depressive symptoms during pregnancy, pregnancy related anxiety, parenting stress, and job strain during pregnancy, are at risk for having an excessively crying infant. Women who have several of these antenatal risk factors are at particular risk.[47] A relatively small prospective German study found that mothers with high scores of negative life changes, assessed by means of the Life Experiences Survey (LES) have infants who cry excessively/fuss more often during their first half year of life.[48] Rautava showed prospectively that experience of stress and physical symptoms during pregnancy, dissatisfaction with the sexual relationship, and negative experiences during childbirth were associated with the development of excessive crying.[49] In a retrospective study, Høgdall found a higher risk of infant crying in a subjectively troubled pregnancy.[50]

In a study by Zuckerman, pregnant women who had a higher score on the Centre for Epidemiological Studies – Depression (CES–D) questionnaire for depressive symptoms were more likely to have an infant (aged 8-72 hours) that would be inconsolable or cry excessively.[51] Infants of mothers with increased depressive symptoms cried more per day compared to non-depressed mothers, although a causal relationship could not be found[52,53]. A higher risk for having a crying infant at 5 weeks of age was also prospectively found in very young women, women who did not live with the father, and women with high trait anxiety.[54] Søndergaard showed in a large prospective cohort study that mothers who reported distress during pregnancy had a threefold risk of their infant crying, and almost a twofold risk of infant crying was found for the women who scored high on the General Health Questionnaire (GHQ), and the psychological distress scale, filled out during pregnancy (based on the Symptoms Check List (SLC–90).[55] A reasonable explanation for this phenomenon is the fact that already existing problems will not be solved by the birth of a child.

Consequences and long-term effects on development

Excessive crying might elicit risky behaviour in caregivers: in the Netherlands half of the 7% of parents who place their infant prone to sleep do so because their infant cries excessively or is a bad sleeper.[56] Other consequences of excessive crying may be breastfeeding failure, attachment problems, marital stress, and postpartum depression.[57] In a Dutch study, Reijneveld showed that 5.6% of parents of 6-month old infants reported having smothered, slapped, or shaken their infant at least once because of its crying, while parents who live in highly urbanized areas even reported 13%.[58] The risk of taking these detrimental actions is highest for parents from non-industrialized countries, those with either no job or a job of less than 16 working hours, and those who judged their infant's crying as excessive. Family related factors

such as child rearing problems, restrictive child rearing style, divorce, social isolation, poverty, low level of education, and parental factors such as illness, addiction, low self-esteem, and history of trauma possibly resulting in child abuse have been described as associated with infant crying.[59]
Shaken Baby Syndrome (SBS) is one of the most severe forms of child abuse and might lead to severe handicap or even death. The average age of the victims is 5 to 9 months, and almost all are less than 39 months of age.[60]

The Dutch prevalence of SBS is currently based on an estimation of the Dutch Forensic Institute, Forum Educatief, Centre for Forensic, Medical and Behavioural sciences, and the Dutch Paediatric Surveillance System (NSCK). With 180.000 live births per year, an estimated 60-70 infants would be the victim of a severe form of SBS. These figures are similar to the Dias' study (41.5 cases per 100.000 live births)[60] and British epidemiological studies[61] with 24.6 per 100.000 infants under the age of 12 months. These figures might even be an underestimation, as a retrospective American study showed that in 31.2% of 173 abused infants with head injuries the diagnosis SBS was not recognized.[62] Of the victims, one-third died after the abuse and about 30-50% had severe neurological impairments and physical damage.[63]
Crying is the initiating stimulus reported in many cases of infant abuse and infanticide.[64,65] In as much as 80% of the cases of abuse of a young infant, crying preceded the abuse.[65] Dias showed that in ¾ of all cases of abuse of a baby, parents are responsible for the abuse (37% (step) fathers, 21% mother's boyfriend, and 15% mother).[60]
For a substantial majority of infants with excessive crying, this disappears after the first three months of life.[66] For a subgroup of infants and parents, especially those with substantial additional risk factors, early excessive crying may not resolve, but evolve into a more generalized 'persistent mother-infants distress' syndrome.[66,67] A few other studies investigated long-term detrimental effects of excessive crying on development. Overall findings are, that infants who continue to cry excessively after the age of 4 – 6 months have poorer fine motor abilities, lower IQ scores, a higher prevalence of eating and sleeping difficulties, hyperactivity, discipline problems, and temper tantrums.[68-72]

Interventions

There is no unambiguous intervention for excessive crying, although many have been described. Three systematic reviews show insufficient evidence for simethicone (an anti-foaming agent to reduce gas bubbles in the stomach), dicyclomine (an anticholinergic drug thought to prevent irritable bowel syndrome), methyl scopolamine (an antispasmodic drug), lactase (an enzyme to digest the milk sugar lactose), soy-based formula, casein hydrolysate milk, low lactose milk, sucrose solution, herbal tea, and behavioural modification.[73-75] Lucassen showed in his study into a subgroup of crying infants in a primary care setting that the use of whey

hydrolysate milk does seem effective.[76] A comparative study into parenting methods and associated infant crying and sleeping was carried out in 2 communities with substantially different approaches to infant care (London, UK, conventional Western care, and Copenhagen, Denmark, "infant-demand" care). A third group consisted of London and Copenhagen parents who had decided before birth to adopt proximal infant care, defined as parenting that involved holding the infant ≥ 80% between 8 AM and 8 PM (shared between caregivers as preferred), frequent breastfeeding and rapid response to infant cries. At 5 weeks of age, the groups did not differ in bouts of unsoothable crying, or in so-called colicky crying.[77] At 12 weeks of age, London infants were more likely to be settled at night, but cried 50% more overall than infants in both other groups. Proximal care infants woke and cried most at night, where Copenhagen infants woke and cried least at 12 weeks of age.[77] A community sample of newborns showed that infants, who have a high number of feeds (> 11/24hr) at the age of one week, have a 2.7 (95% CI 1.5 to 4.8) time higher risk to fail to sleep through the night at the age of 12 weeks.[78] When comparing a behavioural programme to a control group, consisting of mothers who received either an "educational booklet and telephone helpline" or "routine services", infants with a high number of feeds are most likely to sleep through the night when they had received the behavioural programme.[77] No differences were found between the two at risk infant groups in their total 24-hour sleep duration, or in most other behaviours at week 12.

Barr et al. described in a randomised controlled trial that one aspect of behaviour modification, carrying the baby continuously, is ineffective in excessively crying infants in industrialized societies[79], although in normally crying infants, additional carrying seems to reduce cry duration and increase feeding frequency[6]. There is evidence that regularity and stimulus reduction may be an effective strategy in reducing excessive crying.[80-82]

Van den Boom studied neonatal irritability and the development of attachment and describes differences in interactive style between 15 mothers with irritable and 15 mothers with nonirritable infants with consequences for the quality of attachment at the end of the first year, as measured with the 'Strange Situation' procedure.[83] Furthermore, in a group of 100 families with an irritable infant, she showed that a preventive intervention program that is aimed at improving maternal sensitive responsiveness in mothers with irritable infants has the effect that intervention mothers were more responsive, more stimulating, and more controlling of their infant's behaviour when compared to control mothers who did not receive an intervention program. Infants of participating mothers were more sociable, display more self-centered behaviour, explored more, cried less, and were significantly more securely attached than control infants (pre-test at the age of 6 months, post-test at the age of 9 months).

Van IJzendoorn and Hubbard[84,85] replicated and extended the Baltimore study of Bell and Ainsworth's[86] in a longitudinal investigation on maternal responsiveness, infant crying and infant attachment security. Fifty families were observed at home during more than 20 hours, and infant crying behaviour as well as maternal responses were

recorded. Mothers and their infants were observed in the 'Strange Situation' procedure at 15 months of age. The Strange Situation is a laboratory procedure used to assess infant attachment style. Descriptive results showed that infants produced about the same number of crying bouts across the first 40 weeks after birth, but the duration of the bouts decreased by half during this period. The duration of crying peaked in the first nine weeks. The descriptive data were similar to the findings of Bell and Ainsworth[86]. Maternal responsiveness influenced crying behaviour. Contrary to the expectations of the authors, the more frequently mothers ignored their infants' crying bouts in the first nine-week period, the less frequently their infants cried in the following nine-week period, even if intervening variables like earlier crying and synchronous responsiveness were controlled for. The authors conclude that 'benign neglect' of fussing may stimulate the emergent abilities in infants to cope with mild distress.[84]
Extending the earlier report on this investigation[85], they found that crying at home did not differentiate between secure and insecure attachment classifications, and it was not related to Strange Situation crying. Mothers of avoidant infants responded most promptly to their infants' crying. The failure to replicate the Baltimore findings was interpreted in terms of 'differential responsiveness'.

Swaddling

Definition

Swaddling is an ancient infant care practice and a form of infant restraint with many variations, both in type and tightness of wrappings used and in start and duration of swaddling. The swaddling-techniques vary from wrapping the infant in bands (the European way) to tightly folding blankets or a sheet around the child (as practiced for example in South America or in the former Soviet Union). In Indian populations, sometimes a cradle or board is used to swaddle the infant on.[87] Research in experimental settings demonstrates that swaddling influences the arousal function, and promotes sleep.[88,89]
Swaddling was an almost universal infant care practise before the 18th century.[90] Swaddling is still common in some countries in the Middle East and South America, and is gaining popularity in the United Kingdom, the United States and the Netherlands.[57,91,92]

Prevalence

In the Netherlands, swaddling is gaining popularity because of its presumed effect to curb excessive crying in infants. A search into an international ethnographic database, including information on child care practices from 53 different societies, found information on 19 of these 53 societies who used some form of infant wrapping

or swaddling, sometimes with use of cradle-boards.[93] In several societies, infants wear these swaddling clothes all day and night.
In 1999, a Dutch study into infant care practices showed that swaddling was used mainly by Turkish (10.9%) and Moroccan parents (14.8%) and less by Dutch parents (6.2%).[94] Almost all Turkish (86.3%) and Moroccan mothers (75.0%) had been swaddled themselves in their infancy. In April 2002, the advice of regularity (sometimes with swaddling added) has been implemented regionally in the south of the Netherlands, where, at the end of 2005, a sample survey showed that regularity was supplemented with swaddling in 7% of all newborns, on average from the age of 6 tot 7 weeks until they were several months olds (personal communication, Gerria Bankers, Vivent). In 2003, a questionnaire study in a Northern region of the Netherlands into the child care practice of fixating of infants found a 3.9% prevalence rate for the use of swaddling or the safe-T-sleep.[95]

Beneficial and detrimental effects

Swaddling an infant might not be without risk. As described in chapter 2, only 9 randomised controlled trials (RCT) that meet the criteria of Cochrane Central Register of Controlled Trials evaluated the effect of swaddling in young infants.[89,96-103]
According to these RCT's, the following beneficial effects of swaddling could be identified:

- less startles and less arousal during sleep, a longer sleep and an increased arousability, and an infant is more likely to return to sleep on his own during REM sleep[89];
- swaddling may reduce the risk of SIDS, because it hinders turning prone[89];
- preterm infants show improved neuromuscular development, less psychological distress, better motor organisation, and more self-regulatory ability when swaddled during weighing[101,102];
- crying is decreased more by swaddling when compared to massage in infants with cerebral damage[99];
- swaddling during heel sticks makes premature infants return to their baseline hearth rate and pulse oxygen saturation measures more quickly, and it can soothe preterm infants after pain[98,103];
- swaddling did not influence breastfeeding parameters such as number and duration of breastfeeds, amount of ingested milk, and total duration of breastfeeding time[96].

Potential adverse effects of swaddling one should be aware of are:

- the development of hip dysplasia;
- the increased risk of overheating;
- an increased risk for Sudden Infant Death Syndrome when the infant is placed in a prone sleeping position or is able to turn prone when swaddled;

- a risk for vitamin D deficiency due to a lack of sunlight;
- acute respiratory infections due to lung compression during swaddling;
- and a delayed weight-loss recovery in infants who are briefly separated from their mothers after birth or are placed in a nursery and receive supplements.

In medical literature, one fatal outcome of swaddling is described in a case report. This concerned a 10-weeks old infant with a fever who was swaddled in several blankets including the head (in a traditional Roma gypsy way) and kept in a heated room.[104] This illustrates that it is very important to use prudence with traditional or newly introduced methods. The danger of implementing what appears to be a good idea without monitoring the effects is amply illustrated by the rise in cot death all across Europe after the traditional infant sleeping position in Europe was changed to the American custom of placing infants prone (1973). The number of lives lost because if this 'good idea' have yet to be estimated.[105] Sudden Infant Death Syndrome (SIDS) was the major cause of infant mortality after the first week of life, accounting for about two thousand infant deaths each year in the European Community and Eastern Europe. Despite large reductions in SIDS mortality rates in many countries in the last few years, SIDS is still a major cause of postperinatal infant mortality and there are still, on average, several such tragedies everyday somewhere in Europe.

A randomised trial into the efficacy of swaddling on excessive crying

Build-up to the study

In 1998, a student from Russia, Marina Hamidzai, participated in a social paediatric education program in the Wilhelmina Children's Hospital (University Medical Centre Utrecht, the Netherlands). Under supervision of professor Schulpen, she performed a pilot study into cot death and cultural differences in infant care practices of Turkish and Moroccan families. This study was a logical consequence of the results of a nationwide study on ethnicity and mortality in children.[106,107] It was found that children from Turkish and Moroccan families had a death rate twice as high as that of native Dutch children. Differences in cot death were even more striking, twice as high for Turkish as for Dutch infants and four times higher than for Moroccan infants. Further research in infant care practices was needed to explain the ethnic differences.[108]
During this pilot study, Hamidzai showed interest in the ancient practice of swaddling, which was familiar to her because of her own cultural background. In this pilot study, 22 Turkish and 44 Moroccan families participated.[109] In 13.7% of the Turkish families and 29.5% of the Moroccan infants, the infants were swaddled. Furthermore, when sleeping, 54.6% of the Turkish and 38.5% of the Moroccan babies wore a baby sleeping sack, 4.5% of the Turkish and 20.4% of the Moroccan

babies wore a hat, 14.9% of the Turkish babies and 7.6% of the Moroccan babies slept in the prone position, and the heater was on in baby's bedroom in 45.5% of the Turkish families and 24.9% of the Moroccan babies.

Ria Blom, a health care nurse, became intrigued by swaddling in 1994 after meeting a Peruvian mother at the healthcare centre where she worked and she developed experience with swaddling as a method that seemed to reduce restlessness and crying and improve sleep in infants. She developed a holistic approach, where swaddling sometimes was used. She wrote a little booklet for parents to use (In doeken gewikkeld het kindeke, *in Dutch*), in which she included a questionnaire. Data of 136 parent questionnaires gave an impression of the swaddling practices and the effects, as did the pilot study.[109] The main results of this selected descriptive study were as follows. The average age of the babies at moment of start of the intervention swaddling was 13 weeks, and at an average age of 24 weeks, the swaddling had been stopped.[110] In 37%, the infants were swaddled during all sleeping periods; the others only swaddled when the infant had trouble sleeping on its own, or, in perception of the parents, needed the swaddling cloths. Reasons to discontinue swaddling were the infant's protest or resistance, infants breaking loose from the cloths, infants turning prone, warm weather, or because of negative reactions of their youth health care doctor. Turning to the prone position occurred in 21% of all reported babies, their average age was about 7 months. However, the researchers did not obtain data on whether or not the babies were swaddled at the time they were found in bed in the prone position.[110]

The Russian student, Ms Hamidzai, and the health care nurse, Ms Blom, were brought in contact with each other. In a mini focus-group dr. L'Hoir observed their swaddling methods and discussions. L'Hoir's interest in swaddling was threefold. First, from epidemiological studies into the risk factors of cot death, it is known that about half of the parents who still placed infants prone to sleep, which is a risk factor for cot death, did so because of restlessness or crying of their infant.[56] Parents became hopeless and in their fatigue and despair, they placed their infants prone to sleep. Some children fell asleep immediately, slept for a long time, and very deeply. Although it is an understandable reaction this is exactly what cot death prevention workers do not want parents to do. The second reason was fostered by the involvement of L'Hoir with parents of babies who had injuries caused by shaking (Shaken Baby Syndrome). In many of these cases, excessive crying initiated the shaking. The third reason was the fact that the Dutch well baby clinics noticed that swaddling was used more and more often by Dutch parents as a method to curb crying and restlessness, although its efficacy had never been proven. In the Netherlands, well baby clinics prefer to work with evidence-based methods, and, as far as we knew, there were no randomised trials that studied the effect of swaddling on excessive crying.

These three practice based experiences resulted in a grant, written by dr. L'Hoir and dr. Engelberts, which was honoured by ZonMw, the Netherlands Organisation for Health Research and Development.

During the writing of the grant, the most recent booklet of Ms Blom (Rest through *regularity and swaddling, in Dutch*[111]) was evaluated by 2 university students Biology of the Utrecht University with specialism Health Education and Health Promotion (GezondheidsVoorlichting en –Opvoeding, GVO) via a written questionnaire among 95 health nurses and 91 parents. Based on the results of the analyses, the researchers concluded that the overall legibility was good, although the level of difficulty was not consistent. Furthermore, the transfer of information was not optimal and unambiguous and classification of subjects could be improved. With these conclusions, a revised version of the booklet was written in which the content hardly changed, but legibility was improved.[112]

The approach used by Blom[91] was rather similar to the approach as described by Hofacker[81], Wolke[82], and Keefe[80], who demonstrated an effect of regularity and stimulus reduction on crying. All elements of the approach are made congruent to the cot death prevention program.
Studying the effect of swaddling on excessive crying was a challenge. The final goal of the study was to reduce excessive crying and we hypothesized that this might require additional strategies. Regularity and stimulus reduction had already been demonstrated to have some effect in earlier studies.[80-82]
The main question of the randomised trial was whether swaddling had an added effect, next to offering regularity and stimulus reduction.

Aim of the study

The aim of the study was to define if swaddling, when supplementary to regularity and stimulus control, has an added effect on excessive crying. The second goal was to determine in what amount swaddling influences parental perception of their infant's cry.

Outline of the thesis

The **first Chapter** is a brief introduction on normal and excessive crying, swaddling and the period leading up to the randomised trial.
In **Chapter 2**, the possible benefits and detrimental effects of swaddling, based on best evidence in current literature, are described (historical review, effects on sleep and arousal, temperature control, motor development, Sudden Infant Death Syndrome (SIDS), respiratory infections, rickets and Developmental Dysplasia of the Hip (DDH), effects on pain control, effects on crying behaviour, effects on breastfeeding and postnatal weight, and swaddling start and duration).
In **Chapter 3**, differences in infant care practices related to cot death in Turkish, Moroccan and Dutch families in the Netherlands are described. The use of swaddling in the study group as well as estimated prevalence rates of mothers being swaddled during their own childhood is presented.
In **Chapter 4**, the possible effect of adding swaddling compared to a standardized approach of regularity and stimulus reduction to reduce excessive crying is investigated and results of this randomised trial are presented.
In **Chapter 5**, the amount of infants that received a re-challenge, re-elimination test and were really cow's milk allergic diagnosed by an open or double-blind provocation test is investigated. Furthermore, the determinants that might discriminate between favourable and non-favourable reactions to hypoallergenic diet are identified.
In **Chapter 6**, psychological state and social situation of mothers of infants who cried excessively are investigated, and then compared to control mothers of non-excessively crying infants. Furthermore, the relation between high and low maternal scores on several psychological measurements (clinical versus non-clinical range) and the outcome of the intervention is investigated.
In **Chapter 7**, maternal psychological characteristics and their seriousness in relation to excessively crying infants, moderately crying infants and controls is investigated. Furthermore, the effect of two interventions (regularity compared to regularity with swaddling added) was compared between excessively crying infants and moderately crying infants.
In **Chapter 8**, a general discussion and a conclusion on the added effect of swaddling next to regularity on the occurrence of excessive crying is presented. Practical implications are given. Finally, hypotheses, sometimes still somewhat preliminary, on and directions for future research are described.
In **Chapter 9**, an English and Dutch summary is given.
In **Appendix I**, the application of the two support programmes, used in the randomised trial (Chapter 4) is described extensively for youth health nurses.
Although our RCT did not prove added benefit for swaddling, it is often used in the Netherlands, sometimes because of its perceived effect and sometimes because of cultural preference in certain ethnic groups. A safety conscious guideline can be helpful in those instances.

References

1. Ostwald P. The sounds of infancy. Dev Med Child Neurol 1972;14:350-61
2. Zeifman DM. Developmental aspects of crying: infancy, childhood, and beyond. In: Vingerhoets AJJM, Cornelis RR. (Eds.) Adult crying. A biopsychosocial approach. Brunner-Routledge, East Sussex, 2001
3. Brazelton TB. Crying in infancy. Pediatrics 1962;29:579-88
4. Bell SM, Ainsworth MD. Infant crying and maternal responsiveness. Child Dev 1972;43:1171-90
5. Rebelsky F, Black R. Crying in infancy. J Gen Psychol 1972;121:49-57
6. Hunziker UA, Barr RG. Increased carrying reduces infant crying: A randomised controlled trial. Pediatrics 1986:77:641-8
7. Barr RG. The normal crying curve: what do we really know? Dev Med Child Neurol 1990;32:368-72
8. Peiper A. Cerebral function in infancy and childhood. Consultants Bureau, New York, 1963
9. Crying in babies. In: Brugman E, Reijneveld SA, Hollander-Gijsman ME den, Burgmeijer RJF, Radder JJ. Gauging in the child health care 1997-1998 (41-45). Leiden: TNO Prevention and Health, 1999
10. Wessel, MA, Cobb JC, Jackson EB, Harris GS, Detwiler AC. Paroxysmal fussing in infancy, sometimes called "colic". Pediatrics 1954;94:322-32
11. Barr RG. Infant crying and colic: it's a family affair: invited commentary. Inf Ment Health 1995;16:218-20
12. Lehtonen LA, Rautava PT. Infantile colic: natural history and treatment. Curr Probl Pediatr 1996;26:79-85
13. Räihä H, Lehtonen L, Huhtala V, Saleva K, Korvenranta H. Excessively crying infant in the family: mother-infant, father-infant and mother-father interaction. Child Care Health Dev 2002;28:419-29
14. Papoušek M, von Hofacker N. Persistent crying in early infancy: a non-trivial condition of risk for the developing mother-infant relationship. Child Care Health Dev 1998;24:395-424
15. Räihä H, Lehtonen L, Korvenranta H. Family context of infantile colic. Inf Ment Health 1995;16:206-217
16. Barr RG, Rotman A, Yaremko J, Leduc D, Francoeur TE. The crying of infants with colic: a controlled empirical description. Pediatrics 1992;90:14-21
17. Barr RG, Kramer MS, Boisjoy C, McVey-White L, Pless IB. Parental diary of infant cry and fuss behaviour. Arch Dis Child 1988;63:380-7
18. Lester BM, Boukydis CFZ, Garcia-Coll CT, Holl WT, Peucker M. Infantile colic: cry characteristics, maternal perception of cry, and temperament. Infant Behav Dev 1992;15:15-26
19. Reijneveld SA, Brugman E, Hirasing RA. Excessive infant crying: definitions determine risk groups. Arch Dis Child 2002;87:43-4
20. Reijneveld SA, Brugman E, Hirasing RA. Excessive infant crying: the impact of varying definitions. Pediatrics 2001;108:893-7

21. Sleuwen BE van, L'Hoir MP, Engelberts AC, Busschers WB, Westers P, Blom MA, Schulpen TWJ, Kuis W. Comparison of behaviour modification with and without swaddling as interventions for excessive crying. J Pediatr 2006;149:512-7
22. Berg MP van den, Ende J van der, Crijnen AAM, Hofman A, Jaddoe VWV, Mackenbach JP, Moll HA, Tiemeier H, Hengeveld MW, Verhulst FC. Maternal and paternal depression during pregnancy and excessive infant crying. The Generation-R study. Thesis Erasmus University, 2006
23. Wal MF van der, Boom DC van den, Pauw-Plomp H, Jonge GA de. Mothers' report of infant crying and soothing in a multicultural population. Arch Dis Child 1998;79:312-7
24. Canivet C, Hagander B, Jakobsson I, Lanke J. Infantile colic - less common than previously estimated? Acta Paediatrica 1996;85:454-8
25. Alvarez M, St James-Roberts I. Infant fussing and crying patterns in the first year in an urban community in Denmark. Acta Paediatrica 1996;85:463-6
26. St James-Roberts I, Halil T. Infant crying patterns in the first year: normal community and clinical findings. J Child Psychol Psych 1991;32:951-68
27. Forsyth BW, Leventhal JM, McCarthy PL. Mothers' perceptions of problems of feeding and crying behaviour. Am J Dis Child 1985;139:269-72
28. St James-Roberts I. Managing the crying of 1-3 month old infants in western cultures. Presentation conference Paediatric Section of the Royal Society of Medicine, 2002
29. Michelsson K, Rinne A, Paaganen S. Crying, feeding and sleeping patterns in 1 to 12 month-old infants. Child Care Health Dev 1990;16:99-111
30. St James-Roberts I, Hurry J, Bowyer J. Objective confirmation of crying durations in infants referred for excessive crying. Arch Dis Child 1993;68:82-4
31. Zwart P, Vellema-Goud MG, Brand PL. Characteristics of infants admitted to the hospital for persistent colic, and comparison with healthy infants. Acta Peadiatr 2007;96:401-5
32. Lucassen PLBJ, Assendelft WJJ, Gubbels JW, Eijk JTM van, Geldrop WJ van, Knuistingh Neven A. Effectiveness of treatments for infantile colic; systematic review. *BMJ* 1998;316:1563-9
33. Barr RG, Gunnar M. Colic: The 'transient responsivity' hypothesis (p 41 - 66). In: Barr RG, Hopkins B, Green JA (Eds.). Crying as a sign, a symptom, and a signal. MacKeith Press, London 2000
34. Gormally S. Clinical clues to organic etiologies in infants with colic. In: R.G. Barr, I. St James-Roberts, M.R. Keefe (eds.). New evidence on unexplained early infant crying: its origins, nature & management (pp. 133-148). New Brunswick, Johnson & Johnson Pediatric Institute, 2001
35. Treem WR. Assessing crying complaints: the interaction with gastroesophageal reflux and cow's milk protein intolerance. In: R.G. Barr, I. St James-Roberts, M.R. Keefe (eds.). New evidence on unexplained early infant crying: its origins, nature & management (pp. 165-176). New Brunswick, Johnson & Johnson Pediatric Institute, 2001

36. Forsyth BWC, McCarthy PL, Leventhal JM. Problems of early infancy, formula changes, and mothers' beliefs about their infants. J Pediatr 1985;106:1012-17
37. Breslow L. A clinical approach to infantile colic: a review of ninety cases. J Pediatr 1957;50:196-206
38. Alvarez M. Caregiving and early infant crying in a Danish community. J Dev Behav Pediatr 2004;25:91-8
39. St James-Roberts I. Conroy S, Wilsher K. Links between maternal care and persistent infant crying in the early months. Child Care Health Dev 1998;24:353-76
40. White BP, Gunnar MR, Larson MC, Donzella B, Barr RG. Behavioral and physiological responsivity, sleep, and patterns of daily cortisol production in infants with and without colic. Child Dev 2000;71:862-77
41. St James-Roberts I, Conroy S, Wilsher K. Clinical, developmental and social aspects of infant crying and colic. Early Dev Parenting 1995;4:177-89
42. Checkley S. The neuroendocrinology of depression and chronic stress. Br Med Bull 1996;52:597-617
43. Hong HJ, Shin DW, Lee EH, et al. Hypothalamic-pituitary-adrenal reactivity in boys with attention deficit hyperactivity disorder. Yonsei Med J 2003;44:608-14
44. King JA, Barkley RA, Barret S. Attention-deficit hyperactivity disorder and the stress response. Biol Psychiatry 1998;44:72-4
45. Lundly BL, Jones NA, Field T, et al. Prenatal depression effects on neonates. Infant Behav Dev 1999;22:119-29
46. O'Connor TG, Heron J, Glover V. Antenatal anxiety predicts child behavioural/emotional problems independently of postnatal depression. J Am Acad Child Adolesc Psychiatry 2002;41:1470-7
47. Wal MF van der, Eijsden M van, Bonsel GJ. Stress and emotional problems during pregnancy and excessive infant crying. J Dev Behav Pediatr 2007;28:431-7
48. Wurmser H, Rieger M, Domogalla C, Kahnt A, Buchwald J, Kowatsch M, Kuehnert N, Buske-Kirschbaum A, Papoušek M, Pirke K, von Voss H. Association between life stress during pregnancy and infant crying in the first six months postpartum: a prospective longitudinal study. Early Hum Dev 2006;82:341-9
49. Rautava P, Helenius H, Lehtonen L. Psychosocial predisposing factors for infantile colic. BMJ 1993;307:600-4
50. Høgdall CK, Vestermark V, Birch M, Plenov G, Toftager-Larsen K. The significance of pregnancy, delivery and postpartum factors for the development of infantile colic. J Perin Med 1991;19:251-7
51. Zuckerman B, Bauchner H, Parker S, Cabral H. Maternal depressive symptoms during pregnancy, and newborn irritability. J Dev Behav Pediatr 1990;11:190-4
52. Milgrom J, Westley DT, McCloud PI. Do infants of depressed mothers cry more than other infants? J Paediatr Child Health 1995;31:218-21

53. Miller AR, Barr RG, Eaton WO. Crying and motor behaviour of six-week-old infants and postpartum maternal mood. Pediatrics 1993;92:551-8
54. Canivet CA, Östergren PO, Rosén A, Jakobsson I, Hagander BM. Infantile colic and the role of trait anxiety during pregnancy in relation to psychosocial and socioeconomic factors. Scan J Public Health 2005;33:26-34
55. Søndergaard C, Olsen J, Friis-Hasche E, Dirdal M, Thrane N, Sorensen HT. Psychosocial distress during pregnancy and the risk of infantile colic: a follow-up study. Acta Paediatrica 2003;92:811-6
56. Jonge GA de, L'Hoir MP, Ruys JH, Semmekrot BA. Cot death, experiences and insights [In Dutch]. Noorden: Dutch Cot Death Foundation, 2002
57. Karp H. The happiest baby on the block. London: the Penguin Group, 2002
58. Reijneveld SA, Wal MF van der, Brugman E, Hira Sing R, Verloove-Vanhorick SP. Infant crying and abuse. Lancet 2004;364:1340-2
59. Pauli-Pott U, Becker K, Mertesacker T, Beckmann D. Infants with "colic"-mothers' perspectives on the crying problem. J Psychosomatic Research 2000;48:125-32
60. Dias MS, Smith K, deGuehery K, Mazur P, Li V, Shaffer ML. Preventing abusive head trauma among infants and young children: A hospital-based, parent education program. Pediatrics 2005;115:470-7
61. Barlow KM, Minns RA. Annual incidence of shaken impact syndrome in young children. Lancet 2000;356:1571-2
62. Jenny C, Hymel KP, Ritzen A, Reinert SE, Hay TC. Analysis of missed cases of abusive head trauma. JAMA 1999;281:621-6
63. Hennes H, Kini N, Palusci VJ. The epidemiology, clinical characteristics, and public health implications of shaken baby syndrome. J Agression Maltreatment Trauma 2001;5:19-39
64. Frodi A. When empathy fails: aversive infant crying and child abuse. In: Lester BM & Boukydis CFZ (Eds.). Infant crying. Theoretical and research perspectives. Plenum Press: New York, 1985
65. Weston J. The pathology of child abuse. In: Helfer R, Kempe C (Eds.). The Battered Child. Chicago: University of Chicago Press, 1968
66. Barr RG. Crying in the first year of life: good news in the midst of distress. Child Care Health Dev 1998;24:425-39
67. Papoušek M, von Hofacker N. Persistent crying in early infancy: a non-trivial condition of risk for the developing mother-infant relationship. Child Care Health Dev 1998;24:395-424
68. Kries R von, Kalies H, Papoušek M. Excessive crying beyond 3 months may herald other features of multiple regulatory problems. Arch Pediatr Adolesc Med 2006;160:508-11
69. Wake M, Morton-Allen E, Poulakis Z, Hiscock H, Gallagher S, Oberklaid F. Prevalence, stability, and outcomes of cry-fuss and sleep problems in the first 2 years of life: prospective community-based study. Pediatrics 2006;117:836-42

70. Rao MR, Brenner RA, Schisterman EF, Vik T, Mills JL. Long term cognitive development in children with prolonged crying. Arch Dis Child 2004;89:989-92
71. Wolke D, Rizzo P, Woods S. Persistent infant crying and hyperactivity problems in middle childhood. Pediatrics 2002;109:1054-60
72. Rautava P, Lehtonen L, Helenius H, Silanpaa M. Infantile colic and family three years later. Pediatrics 1995;95:43-7
73. Wade S, Kilgour T. Extracts from "Clinical Evidence": Infantile colic. BMJ 2001;323:437-40
74. Garrison MM, Christakis DA. A systematic review of treatments for infantile colic. Pediatrics 2000;106:184-90
75. Lucassen PLBJ, Assendelft WJJ, Gubbels JW, Eijk JTM van, Geldrop WJ van, Knuistingh Neven A. Effectiveness of treatments for infantile colic; systematic review. BMJ 1998;316:1563-69
76. Lucassen LB, Assendelft WJ, Gubbels LW, Eijk JT van, Douwes AC. Infantile colic: crying time reduction with a whey hydrolysate; a double blind, randomized placebo-controlled trial. Pediatrics 2000;106:1349-54
77. St James-Roberts I, Alvarez M, Csipke E, Abramsky T, Goodwin J, Sorgenfrei E. Infant crying and sleeping in London, Copenhagen and when parents adopt a "proximal" form of care. Pediatrics 2006;117:1146-55
78. Nikolopoulou M, St James-Roberts I. Preventing sleep problems in infants who are at risk of developing them. Arch Dis Child 2003;88:108-11
79. Barr RG, McMullan SJ, Spiess H, Leduc DG, Yaremko J, Barfield R, et al. Carrying as colic 'therapy': a randomized controlled trial. Pediatrics 1991;87:623-30
80. Keefe MR, Barbosa GA, Froese-Fretz A, Kotzer AM, Lobo M. An intervention program for families with irritable infants. MCN 2005;30:230-6
81. Hofacker N von, Papousek M, Jacubeit T, Malinowski M. Rätsel der Säuglingskoliken. Ergebnisse, Erfahrungen und therapeutische Interventionen aus der "Müncher Sprechstunde für Schreibabies". Monatsschr Kinderheilkd 1999;147:244-53
82. Wolke D, Gray P, Meyer R. Excessive infant crying: a controlled study of mothers helping mothers. Pediatrics 1994;94:322-32
83. Boom DC van den. Neonatal irritability and the development of attachment: observation and intervention. Thesis, Rijksuniversiteit Leiden, 1988
84. IJzendoorn MH van, Hubbard FOA. Are infant crying and maternal responsiveness during the first year related to infant-mother attachment at 15 months? Attach Hum Dev 2000;2:371-91
85. Hubbard FOA, IJzendoorn MH van. Maternal unresponsiveness and infant crying across the first 9 months: A natural longitudinal study. Infant Behav Dev 1991;14:299-312
86. Bell SM, Ainsworth MDS. Infant crying and maternal responsiveness. Child Dev 1972;43:1171-90
87. Chisholm JS. Swaddling, cradleboards and the development of children. Early Hum Dev 1978;2/3:255-75

88. Franco P, Seret N, Hees JN van, Scaillet S, Groswasser J, Kahn A. Influence of swaddling on sleep and arousal characteristics of healthy infants. Pediatrics 2005;115:1307-11
89. Gerard CM, Harris KA, Thach BT. Spontaneous arousals in supine infants while swaddled and unswaddled during rapid eye movement and quiet sleep. Pediatrics 2002;110:e70
90. Lipton EL, Steinschneider A, Richmond JB. Swaddling, a child care practice: historical, cultural and experimental observations. Pediatrics 1965;35(suppl):521-67
91. Blom MA. Crying and Restlessness in Babies. A Parent's Guide to Natural Sleeping Edinburgh: Floris Books, 2005
92. Masataka N. On the function of swaddling as traditional infant-care practiced by native South Americans. Shinrigaku Kenkyu 1996;67:285-91
93. Nelson EAS, Schiefenhoevel W, Haimerl F. Child care practices in nonindustrialized societies. Pediatrics 2000;105:e75
94. Sleuwen BE van, L'Hoir MP, Engelberts AC, Westers P, Schulpen TWJ. Infant care practices related to cot death in Turkish and Moroccan families in the Netherlands. Arch Dis Child 2003;88:784-8
95. Thuiszorg De Friese Wouden. Enquête vastleggen van kinderen 2004. Thuiszorg De Friese Wouden, 2004
96. Bystrova K, Matthiesen AS, Widström AM, Ransjö-Arvidson AB, Welles-Nyström B, Voroonstov I, Uvnäs-Moberg K. The effect of Russian Maternity Home routines on breastfeeding and neonatal weight loss with special reference to swaddling. Early Hum Dev 2007;83:29-39
97. Tsogt B, Maniseki-Holland S, Pollock J, Blair P, Fleming P. Can traditional care influence thermoregulation: A prospective controlled study of the effects of swaddling on infants' thermal balance in a Mongolian winter. The 9th SIDS Conference, Yokohama, Japan, June 1-4 2006
98. Huang CM, Tung WS, Kuo LL, Ying-Ju C. Comparison of pain responses of premature infants to the heelstick between containment and swaddling. J Nurs Res 2004;12:31-40
99. Ohgi S, Akiyama T, Arisawa K, Shigermori K. Randomised controlled trial of swaddling versus massage in the management of excessive crying in infants with cerebral injuries. Arch Dis Child 2004;89:212-216
100. Short MA. A comparison of temperature in VLBW infants swaddled versus unswaddled in a double-wall incubator in skin control mode. Neonatal Network 1998;17:25-31
101. Neu M, Browne JV. Infant physiologic and behavioral organization during swaddled versus unswaddled weighing. J Perinat 1997;17:193-198
102. Short MA, Brooks-Brunn JA, Reeves DS, Yeager J, Thorpe JA. The effects of swaddling versus standard positioning on neuromuscular development in very low birth weight infants. Neonatal Network 1996;15:25-31
103. Campos RG. Soothing pain-elicted distress in infants with swaddling and pacifiers. Child Dev 1989;60:781-792

104. Gestel JPJ van, L'Hoir MP, Berge M ten, Jansen NJG, Plötz FB. Risks of ancient practices in modern times. Pediatrics 2002;110:e78
105. Carpenter RG, England PD. Report of the European Concerted Action on SIDS (p 12). Department of Epidemiology and Population Sciences, London School of Hygiene and Tropical Medicine, London, 1998
106. Steenbergen JE van, Schulpen TWJ, Hoogenboezem J, Driel HF van, Bijlsma F. Ethnicity and Childhood Mortality in the Netherlands. Eur J of Public Health 1999;9:205-10
107. Schulpen TWJ, Steenbergen JE van, Driel HF van. Influences of ethnicity on perinatal and child mortality in the Netherlands. Arch Dis Child 2001;84:222-6
108. Sleuwen BE van, L'Hoir MP, Engelberts AC, Westers P, Schulpen TWJ. Infant care practices related to cot death in Turkish and Moroccan families in the Netherlands. Arch Dis Child 2003;88:784-8
109. Hamidzai M. Cot death and differences in infant care practices in Turkish and Moroccan families. A pilot study. Utrecht University, 1998
110. Beltman M. Swaddling of infants. An old fashioned habit on its return. Literature review. Utrecht University, 2000
111. Blom R. Rest through Regularity and Swaddling [In Dutch]. A Parent's Guide. Utrecht: De Bakermat, 2001
112. Bodegraven A van, Postelmans K. Evaluation of the booklet 'Rest through Regularity and Swaddling. A parent's Guide.' Utrecht University, 2001

Swaddling – A systematic review

2

Bregje E. van Sleuwen
Adèle C. Engelberts
Magda M. Boere-Boonekamp
Wietse Kuis
Tom W.J. Schulpen
Monique P. L'Hoir

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Abstract

Swaddling was an almost universal child-care practice before the 18th century. It is still tradition in certain parts of the Middle East and is gaining popularity in the United Kingdom, the United States, and the Netherlands to curb excessive crying. We have systematically reviewed all articles on swaddling to evaluate its possible benefits and disadvantages. In general, swaddled infants arouse less and sleep longer. Preterm infants have shown improved neuromuscular development, less physiologic distress, better motor organization, and more self-regulatory ability when they are swaddled. When compared with massage, excessively crying infants cried less when swaddled, and swaddling can soothe pain in infants. It is supportive in cases of neonatal abstinence syndrome and infants with neonatal cerebral lesions. It can be helpful in regulating temperature but can also cause hyperthermia when misapplied. Another possible adverse effect is an increased risk of the development of hip dysplasia, which is related to swaddling with the legs in extension and adduction. Although swaddling promotes the favourable supine position, the combination of swaddling with prone position increases the risk of sudden infant death syndrome, which makes it necessary to warn parents to stop swaddling if infants attempt to turn. There is some evidence that there is a higher risk of respiratory infections related to the tightness of swaddling. Furthermore, swaddling does not influence rickets onset or bone properties. Swaddling immediately after birth can cause delayed postnatal weight gain under certain conditions, but does not seem to influence breastfeeding parameters.

Key Words

swaddling, systematic review, traditional care, sudden infant death syndrome, developmental dysplasia of the hip

Some degree of infant restraint, known as swaddling (also called binding or bundling), with or without the use of a cradleboard was an almost universal childcare practice before the 18th century.[1] Swaddling is still common in some countries in the Middle East and South America and is gaining popularity in the United Kingdom, the United States, and the Netherlands.[2–4] In this review, all known studies of swaddling were included. Most studies were descriptive and not randomised, uncontrolled, or comparative. Many results, therefore, comprised opinions and perceptions. The 9 randomised comparative studies of good quality will be discussed separately.
A historical overview of swaddling and its practice in relation to sleep state and arousal, temperature control, motor development, sudden infant death syndrome (SIDS), respiratory infections, bone development, developmental dysplasia of the hip (DDH), pain control, crying behaviour, breastfeeding, and weight gain after birth and the duration of swaddling will be described.

Historical review

The technique of swaddling varies from wrapping the infant in bands (the European way) to tightly folding blankets or sheets around the child (as practiced for example in South America and in countries of the former Soviet Union). Swaddling is of considerable interest, because the use of this practice is widely spread in many different societies. The Holy Bible describes infants winded in cloths, and one of the earliest illustrations of swaddling is of the infant Jesus.[5] A few centuries ago swaddling was used in most societies of the north temperate and subarctic regions, in the Mediterranean and Middle East areas, in Asia and South America, and many other parts of the world.[4,6] In 1971 almost 52% of 139 societies still used some form of infant restraint.[7] In 2003 swaddling was still common in the Middle East.[8] In parts of the world where humidity and temperature are high, such as Africa, swaddling can promote skin infections; as an alternative, children are carried in a sling, often with a minimum of clothing.[9] Swaddling already began to disappear in Europe before industrialization. One of the reasons for the decrease of swaddling is that in the 20th century it was confined to a few rural societies in eastern Europe.[1] Rousseau's enormously successful book *Emile*,[10] in which he described swaddling as "unnatural," could have contributed to this decrease. In the 19th and 20th centuries, more "liberal" ideas concerning child rearing started to dominate. Another influence could be the "swaddling hypothesis."[11–14] According to this hypothesis, the restraint of swaddling leads to an adult personality structure that inclines people to alternate meek submission and ambivalently regarded authority, with explosive, excessive expression of emotion. "Adult Russians and Eastern Europeans who were swaddled, learned that passivity and restraint are necessary to secure milk, love and freedom, which can then only be enjoyed in excessive outbursts of emotion which are ambivalent nonetheless, for the mother is both the cause of 'imprisonment' and the release from it."[11,13] This hypothesis was never really defined or tested but probably reflects the sentiments concerning swaddling in European society at the time.

Swaddling is still applied in various traditional societies, sometimes using a board or cradle.[6] However, in societies where it was virtually abandoned, there seems to be a revival of swaddling practices because of presumed favourable effects on infant behaviour, especially its soothing effect on infants' crying.[2,3] One should realize that there is not one method of swaddling but many variations both in start and duration of swaddling and type and tightness of wrappings used. Apart from its presumed benefits, there are serious considerations to be addressed concerning its possible detrimental effects, specifically in relation to the congenital dislocation of the hip, the development of acute respiratory infections, and the combination of swaddling with prone position for the risk of SIDS.[15–17] In the Netherlands, swaddling has rapidly gained popularity in the last 10 years, mainly because it is presumed that this traditional method curbs excessive crying in infants. Nowadays, ~8% of Dutch parents place their infants in the prone position to sleep.[18] Half of these parents do so because their infant cries excessively or sleeps poorly. It was shown in 1 study that in 80% of the cases of child battering, excessive crying preceded the violence.[19] Because in the last few years many reports were published on the effects of swaddling, we decided to conduct a systematic review on the effects of swaddling infants.

Methods

Search Strategy

Electronic searches were conducted in PubMed (1966 to February 2007), PsycINFO (1887 to February 2007), Embase (1974 to February 2007), the Cochrane library (2007, Issue 1), and Blackwell Synergy (1990 to February 2007). We used the Medical Subject Headings (MeSH) heading swaddling. Manually searched reference lists were used also.

Selection of Eligible Trials

All published randomised, controlled trials (RCTs) that evaluated the intervention of swaddling were included, as were all other studies on swaddling in relation to sleep state and arousal, temperature control, motor development, SIDS, (acute) respiratory infections, bone development, DDH, pain control, the effect on crying behaviour, and breastfeeding and neonatal weight loss.

Results

Among the 78 articles reviewed, we identified 9 RCTs that met the criteria of the Cochrane Central Register of Controlled Trials (Table 1).[20–28] Of all articles reviewed, 9 were articles on swaddling in relation to sleep state and arousal, 8 articles described the effect of swaddling on temperature control, 6 articles discussed the effect of swaddling on motor development, 12 articles evaluated the risk of SIDS related to swaddling, 5 set out the relation between swaddling and the risk for (acute) respiratory infections, 1 article was on the relation between swaddling and rickets and bone properties, 14 articles involved swaddling-related DDH, 4 articles set out the effects of swaddling on pain control, and the effect on crying behaviour was discussed in 4 articles. The effect on breastfeeding and neonatal weight loss was discussed in 1 article, and the mean duration of swaddling in different cultures was described in 5 articles. All articles reviewed, including the Cochrane RCTs, will be described according to subject.

Table 1. Characteristics of Cochrane RCTs

	Gerard, 2002[20]	Short, 1998[25]	Neu, 1997[22]	Short, 1996[21]	Campos, 1989[24]	Ohgi, 2004[23]	Huang, 2004[26]	Tsogt, 2006[27]
Interventions	Swaddled v unswaddled	Swaddled v unswaddled	Swaddled v unswaddled	Swaddling v standard positioning	Pacifier v swaddling	Swaddling v massage	Swaddling v containment	Swaddling v infant sleeping bag
No of infants	26 (16 female, 10 male)	15 VLBW infants	14 preterm infants	50 VLBW infants (25 female, 25 male)	64 (32 at 2 weeks, 32 at 2 months)	25 (13 swaddled, 12 massage)	32 premature infants, gestational age <37 weeks	80 (40 swaddled, 40 infant sleeping bag)
Study duration	107 ± 7 min, single event during daytime nap	-	-	Swaddling 21.1 hours a day, control group 1.6 hours a day	Single event at 2-weeks old and 2-months old	Three weeks, swaddling 3.4 times a day, massage 3.3 times a day	5 minutes before to 11 minutes after heelstick	Periods of 24 hour at ages 1 and 3 months
Mean age	80 ± 7 days	29.1 ± 1.5 weeks	32 weeks gestational age	28 weeks gestational age	2 weeks and 2 months	11 weeks	-	1 month and 3 months
Effect size	- No change in sighs (in QS and REM sleep) - Less startles during QS (p<.02) and REM sleep (p<.005)decrease in behavioral arousals during QS (p<.001) - Decrease of progressing sights to startles in QS and REM sleep (p<.003 and 0.0004) - Decrease of progression startles to full arousal in QS (p<.006) - Increase in brief arousals during REM sleep (p<.05) - Increase in duration in REM sleep (p<.0005)	When swaddled: - Abdominal temperature 0.2 degree C higher - Lower incubator temperature needed (mean = 26.9 ± 0.4) than when unswaddled (mean = 29.8 ± 0.5)	When swaddled: - Less physiologic distress (p<.002) - Better motor organization (p<.001) - More effective self-regulatory ability (p<.037)	When swaddled: - Higher score on tone and motor subscale (p=.03) - Higher score on response subscale (p=.04) - Higher on total score (p=.007)	At 2 weeks: - HR levels and crying declined more rapidly with pacifier At 2 months: - Similar rates of decline in HR and crying At both ages: - In pacifier group more time in alert state, HR and crying tended to rebound more in pacifier group	When swaddled: - Significant decrease in amount of crying - Post intervention significant improvement in infant behavioral profiles and maternal anxiety level - Parents more satisfied with the effectiveness of intervention	When swaddled: - Significant shorter time periods to return to baseline HR and SpO_2	- No differences in infant core, peripheral and micro-environmental temperature
Level of evidence[A]	2b	2b	2b	2b	2b	2b	2b	2b

A Level of evidence as defined by the Oxford Centre for Evidence-based Medicine Levels of Evidence (May 2001)

Effects on Sleep and Arousal

It is widely held that swaddling calms infants and induces sleep.[6] Several studies have been conducted on the effect of swaddling on sleep state and arousal. Swaddling above the waist seems to increase daytime sleep periods and the total daytime sleep.[29] Lipton et al described in their case-control study1 an increase in sleep, reduced levels of motor activity, fewer startles, and lower heart rate (HR) variability compared with nonswaddled infants. There was no decrease in the capacity to respond to stimulation; there was only a decrease in frequency of response to stimulation. A large ethnological observational study among Navajo infants revealed a definite correlation between cradleboard use and lowered behavioural states.[30] Being swaddled on a cradleboard completely immobilizes an infant and presumably regulates the infant's level of physiologic arousal or responsiveness. In a short-term laboratory nap study (Cochrane, clinical trial), swaddled infants had less startles during quiet sleep (QS) and rapid eye movement (REM) sleep, and a decrease in behavioural arousals during QS but not during REM sleep (Table 1).[20] Swaddling decreased the progression of sighs to startles in both QS and REM sleep, but the progression from startles to full arousal was decreased in QS but not in REM sleep. The percentage of brief arousals was increased in REM sleep during swaddled periods, whereas the average sleep duration when swaddled was increased. In QS, however, there was no difference in either brief arousals or average sleep duration. Several case-control studies were conducted on swaddling and arousal. Swaddling and sleeping supine promotes a better efficiency of sleep, more QS, and fewer spontaneous awakenings compared with sleeping supine but unswaddled.[31] However, when sleeping swaddled or restrained, less-intense auditory stimuli are needed to arouse the infants compared with the infants who where not swaddled.[32] This may explain the increase in brief arousals during REM sleep reported in the previously mentioned study. When sleeping supine, restrained infants have a significant greater and earlier decrease in HR to white-noise challenges than nonswaddled supine infants during REM sleep.[33] During QS, the respiratory rate increases slightly with swaddle tightness, whereas HR, inspiratory tidal volumes, relative minute volume, and pulse oxygen saturation (SpO_2) do not change.[34] On the other hand, during REM sleep, there is a slight decrease in HR after tightening, whereas the respiratory rate, inspiratory tidal volume, volume, and SpO_2 did not show a significant difference. In a study in which swaddling was compared with a free condition, swaddling resulted in less arousal, less alert activity, more drowsiness, and more sleep.[35] Overall, it is clear that swaddling stimulates sleep continuity, as shown under laboratory conditions and in descriptive studies. The effect is most consistent in periods of QS but not always consistent during REM sleep. Swaddling seems to inhibit each step from sighs through startles to full arousal in the arousal pathway. The restraint of the arms may inhibit the movements that are associated with a full extensor startle response. Prone sleeping probably also has this effect; it is associated with fewer arousals and better sleep than those in the supine position.[20]

Swaddling a supine infant, therefore, seems to mitigate these differences. The difference in the physiologic effect of swaddling in QS and REM sleep need more research, however.

Temperature Control

Many different populations have used swaddling to keep infants warm and comfortable.[1,27,36–38] Preliminary results of an RCT (Cochrane RCT) that compared the effects of swaddling (within 48 hours of birth) with use of an infant sleeping bag of equal thermal resistance suggested that infant core, peripheral, and microenvironmental temperature were no different in Gers (traditional circular single-room tents) or apartments, and no differences were found between swaddled and nonswaddled infants.[27] A case-control study (Cochrane RCT) showed that in very low birth weight (VLBW) infants in a heated double-walled incubator, abdominal temperature rose 0.2°C higher during swaddling, and infants required a lower incubator temperature.[25] Hyperthermia, however, could be a possible adverse effect of swaddling, especially when the head is also covered or in case of infection.[37–40] In summary, there is evidence that swaddling can be favourable for temperature control, but there is a risk for hyperthermia when misapplied.

Motor Development

All different techniques of swaddling have in common that they restrict the movement of the body and limbs.[6] In VLBW infants, 1 case-control study (Cochrane RCT) showed that swaddling might have a positive effect on neuromuscular development,[21] and another case-control study (Cochrane RCT) showed that if preterm infants are weighed when swaddled, they show better motor organization.[22] While swaddled, the level of motor activity is reduced.[1] No evidence that swaddling has short-term or long-term effects on the attainment of motor milestones has been found.[6,39,40] Dennis and Dennis[41] found that Hopi infants reared on a cradleboard walked as early as unrestrained Hopi infants.[41–43] Next to the variation in the tightness of swaddling, especially of the lower limbs, most studies have not reported the exact duration of swaddling, which makes it difficult to draw firm conclusions about the effect of swaddling on motor development.

Sudden Infant Death Syndrome

Six articles evaluated swaddling and the risk of SIDS. A large nationwide case-control study (New Zealand) and a prospective cohort study both indicated that firm tucking, wrapping, or swaddling contributes to a reduced risk of SIDS.[17,44] Swaddled infants in the supine position have a lower risk for SIDS.[17,45,46] Immobilization of infant arms and legs by swaddling might reduce the chances that an infant will cover his or her head and face with bedding, which is a well-established risk factor for SIDS in which overheating and asphyxia may be causal factors.[34]

It can be assumed that infants, when swaddled, are placed supine, as Nelson et al[47] stated in a retrospective descriptive study. The "Back to Sleep" campaign has been successful in decreasing the rate of SIDS, but some infants seem to sleep poorly when they are placed supine. Most infants, however, tolerate the supine position when swaddled, even a considerable number of infants who are accustomed to sleeping prone.[34,48] The physical restraint of swaddling presumably prevents infants from turning prone during sleep before they have gained experience in turning to prone and back again when awake.[34] Because inexperienced prone sleepers are especially at risk for SIDS, this could explain, in part, the reduced risk associated with swaddling.[49] A relatively large case-control study combined with a smaller prospective cohort study showed that if swaddled infants are placed prone, they have a 12-fold increase in risk for SIDS, whereas nonswaddled prone infants have a 3-fold increase in risk.[45] In view of this finding, it is important to note that in a small retrospective cohort study, some swaddled infants were already able to turn prone after 3 months of age.[50] Recommendations concerning swaddling, therefore, should address the difference in SIDS risk associated with supine and prone sleeping. Theoretically, the decreased arousal associated with swaddling could also prevent arousal in life-threatening situations. Epidemiologic evidence clearly shows that being supine and swaddled decreases the SIDS risk more than being supine without swaddling; thus, the motor restraint that prevents the life-threatening situations seems to carry more weight than the decreased arousal as long as the infant remains supine.

Some authors warned, in 2 case-control study reports, against tight wrapping and the possible increased risk for SIDS, because hypothetically, respiratory function could be compromised.[51,52] The authors suggested that firm tucking-in while the infant is in a prone position could push his or her face forcefully into the mattress or pillow.[51]

As 1 case report and some studies have suggested, hyperthermia as a possible adverse effect of swaddling could also increase the risk of SIDS, especially when the infant's head is covered.[53–56] On the other hand, 3 studies indicated that swaddling might be protective against SIDS in cold winters, because swaddled infants cool down less.[46,47,57] Williams et al[58] demonstrated that SIDS is also associated with too little thermal isolation, and this is particularly the case when (nonswaddled) infants are not firmly tucked-in.

Respiratory Infections

In a relatively small retrospective cross-sectional cohort study it was shown that infants in Turkey and China who have been (completely or partially) swaddled for at least 3 months have a 4 times higher incidence of (radiologically confirmed) pneumonia and upper respiratory infections than infants who were unswaddled.[16] A hospital-based retrospective case-control study (with nonmatched controls) indicated that being swaddled while exposed to sunlight carries a risk for subclinical vitamin D deficiency in Indian children, which in turn has been associated with an increased risk

for acute lower respiratory infections.[59] This is also suggested in an expert opinion to be of influence in Mongolia, where it is common to swaddle an infant from head to toe, exposing only the face, for the first few weeks or months of life.[38]
In a small case-control study with 40 neonates with radiologically confirmed moderate pneumonia in Beijing, China, Li et al[60] found that unswaddled infants in the prone position had 18% higher oxygen tension than swaddled supine infants and 12% higher oxygen tension than unswaddled supine infants. The fact that the oxygen saturation level is higher in the prone than in the supine position is supported by a small case-control study by Chaisupamongkollarp et al[61] in which prone positioning in spontaneously breathing infants with pneumonia resulted in a statistically significant increase in mean oxygen saturation. Kahn et al[33] also found indications for cardiorespiratory compromise associated with swaddling, especially tight swaddling. This degree of tightness was less than that imposed by some of the traditional methods studied. Overly tight swaddling could lead to atelectasis (incomplete expansion) and potentially increase susceptibility to infection.[34] In conclusion, the tightness of swaddling might influence the risk of respiratory infections. If swaddling causes decreased exposure to sunlight, there is a risk for vitamin D deficiency, which can then increase the risk of respiratory infections. In these studies, however, no confounding factors were included, the groups were small, and some of the methods were inconsistent.

Rickets and DDH

A retrospective case-control study showed that complete or partial swaddling from birth on did not influence the incidence of rickets or bone properties as measured by ultrasound.[62] The term "developmental dysplasia of the hip" (DDH) indicates a dynamic disorder that may get better or worse as an infant develops, depending on management in the first months of life.[63] Eleven epidemiologic studies have shown that the incidence of DDH is highly correlated with the traditional use of swaddling for newborn infants.[15,64–73] In 1 study, other factors such as breech position, congenital muscular torticollis, congenital foot deformities, and positive family history of hip dysplasia were more important risk factors for DDH than postnatal swaddling.[74] Animal models and observations in the neonatal period show that when the hip and knee are forcibly extended either by a diaper or infant clothing, it may cause prolonged tension subluxation or even dislocation of the femoral head.[64,65,75,76] Research into the geographic and racial incidence of DDH shows that in cultures in which the hips are kept in a flexed position, the incidence of DDH is much lower compared with cultures in which the legs are kept in extension (e.g., when swaddled on a cradleboard).[65–67] The incidence of DDH in Japan used to be high before 1965, when a so-called swathing diaper was widely used.[65] In 1973, advice was given in Japan to avoid prolonged extension of the hip and knee of infants during early postnatal life.[67,68]

Afterward, a decrease in the incidence of DDH in 3- to 7-month-old infants was reported. In summary, DDH can be promoted by swaddling, especially when the legs are not free to bend and flex. Attention to this adverse effect is of utmost importance.

Effects on Pain Control

The effect of swaddling on recovery from painful stimuli has been addressed in a few case-control studies. Both pacifiers and swaddling can soothe infants after a pain stimulus (Cochrane RCT).[24] Swaddling is less immediately effective compared with pacifier use but is also less subject to rebound after termination of the intervention compared with a pacifier in term infants.[24] Another RCT (Cochrane RCT) showed that premature infants (<37 weeks' gestational age or <2500 g body weight) in swaddling return to their baseline HR and oxygen saturation (SpO_2) in shorter time periods compared with those in containment (being surrounded by blanket rolls).[26] In preterm infants (postconceptional age: 31–36 weeks), swaddling is an effective means of speeding up recovery from heel lance (decrease in HR, increase in arterial oxygen saturation), whereas in infants of 27 to 30 weeks postconceptional age, recovery after heel lance is not influenced by swaddling, except for an increase in arterial oxygen saturation levels when swaddled.[77]

Effects on Crying Behaviour

As shown in a Cochrane RCT, swaddling in infants with neonatal cerebral insults as an intervention for the management of excessive crying (i.e., at least 3 hours/day of crying for at least 1 week) decreased the amount of crying significantly compared with massage.[23] These infants (aged 0–3 months) were swaddled with a blanket wrapped around the whole body with the head covered and minimal hip restraint over a period of 3 weeks for at least 3 times per day for at least 30 minutes each time. Swaddling was associated with a significant decrease in postintervention crying (measured with a diary that recorded crying, sleeping, and feeding patterns[78,79]) by 28% compared with massage (5%). Furthermore, swaddling was associated with significantly improved neurobehavioral organization (Neonatal Behavioural Assessment Scale cluster scores), a significant reduction of maternal anxiety (State-Trait Anxiety Inventory), and an increase in parental satisfaction after swaddling.
These results suggest that swaddling suppresses crying by protecting infants with brain lesions from external and internal stimulation and improves the neurobehavioral organization of these infants. The reduction in crying that is achieved in this fashion probably helps to alleviate parental anxiety and stress. However, because there have been very few systematic studies of the time-related change in crying in infants with cerebral lesions, longer follow-up studies are needed.

Swaddling can be used as an effective strategy to support infants with neonatal abstinence syndrome.[80] In an RCT, adding swaddling to an intervention that offered regularity and stimulus reduction for excessive crying gave no extra benefit, although a small but significant effect was shown in infants aged 1 to 7 weeks at randomization.[81]

Effects on Breastfeeding and Postnatal Weight

In a Russian randomised trial, 176 mother-infant dyads were studied right after birth of the infant and managed for 5 days. The study showed that infants who were swaddled directly after birth did not differ from infants who were clothed in Western infant attire in respect to breastfeeding variables (number and duration of feeds, amount of supplements, and amount of ingested breast milk).[28] Infants who were swaddled had delayed recovery of weight gain if they were subjected to separation from their mother for the first 2 hours after birth and if they were not able to room in with their mother but were kept in a separate nursery and received supplemental feeds. The authors hypothesized that because touch has been shown to influence growth (e.g., by stimulating the release of gastrointestinal hormones), swaddling could have a negative effect by limiting tactile stimulation. If this hypothesis holds true, the effect of a less-rigorous swaddling might not be as apparent.

Swaddling Start and Duration

In several descriptive studies, the duration of swaddling has been measured, and it seems to vary widely across different cultures. In Chisholm's study into cradleboard use, Navajo Indians place their infants on cradleboards within a few days of birth and continue the use of the board for an average of 10.2 months, ranging up to 2 years of age (J. S. Chisholm, PhD, *Developmental Ethology of the Navajo*, unpublished PhD thesis, 1978). The exact duration is determined individually. The daily amount of time spent on a cradleboard shows wide individual variation but probably averages ~60% to 70% in the first 6 months of life and ~30% later. With the older children, the board may simply be used as a sleeping place (in the Navajo tribe, the cradleboard is also used in an upward position and placed in close proximity of the mother).

A Dutch descriptive retrospective study that included children who were seen at a well-infant clinic where swaddling was integrated in the usual care demonstrated that children were swaddled for an average period of 12 weeks, starting at an average age of 13 weeks.[50] In rural Turkey, swaddling is common practice during the entire first year of life.[29] In the rural minority areas of Yunnan, China, the average duration of swaddling is 35 days.[36] The binding to a cradleboard, accomplished by means of lacing strings or strips of cloth passed over the body and allowing only slight flexion of the legs, is applied to Hopi Indian children in their first 3 months of life.[82] From 3 months until 6 to 12 months the cradle is used only during sleeping or "according to the desires of the child."

A retrospective study showed that Mongolian infants were completely swaddled during their first 4 months of life and partially swaddled until they were 5 months of age, on average.[62] In Russia, it is also common to swaddle infants immediately after birth, leaving only the infant's front face unbound.[28]

Discussion

Only 9 RCTs that evaluated the effect of swaddling were identified.[20–28] The following beneficial effects have been identified.

1. Healthy infants aged 80 ± 7 days (range: 24–180 days) who are swaddled during sleep have less startles, arouse less, and sleep longer.[20] Infants who sleep supine have decreased awakenings during QS when swaddled. During REM sleep, behavioural arousals are unchanged, but an infant is more likely to return to sleep on his or her own. Because prone sleeping is often started when infants sleep poorly or restlessly, these findings are of importance.
2. Preterm infants show improved neuromuscular development when swaddled21 and less physiologic distress, better motor organization, and more self-regulatory ability when swaddled during weighing.[22] In VLBW infants in a double-walled incubator, abdominal temperature is higher when swaddled.[25]
3. In excessively crying infants with cerebral damage, swaddling decreases significantly the amount of crying compared with massage.[23]
4. Swaddling during heel sticks makes premature infants return to their baseline HR and SpO_2 measures more quickly,[26] and it can soothe preterm infants after pain.[24] Swaddling is less-immediately effective than pacifier use but is also less subject to rebound compared with a pacifier.
5. Swaddling did not influence breastfeeding parameters such as number and duration of breastfeeds, amount of ingested milk, and total duration of breastfeeding time.[28]

Before reintroducing this traditional method of infant care, possible detrimental effects need to be addressed. Well-conducted studies have revealed the following potential adverse effects of swaddling.

1. Clear evidence exists about the risk of swaddling for the development of hip dysplasia, especially when the child is swaddled in extension and adduction. This detrimental effect of swaddling is related to the misapplied use of the practice. Swaddling in a manner that allows the hips and knees to move freely might not have this risk-increasing adverse effect, but more studies are needed to test this hypothesis. The studies that showed beneficial effects of swaddling have been conducted with the infants' lower extremities wrapped loosely, but the contribution of wrapping their legs on sleep, pain relief, and excessive crying also needs to be addressed.

2. There have been indications of an increased risk of overheating.
3. There is an increased risk of SIDS, but only when the swaddled infant is placed prone or is able to turn to the prone position
4. Some evidence exists for an added risk of vitamin D deficiency and acute respiratory infections. The relation between swaddling and acute respiratory infections has been set out in 4 studies, all of which were conducted in non-Western countries. In these countries, a higher risk of respiratory infections also seems to be related to the tightness of swaddling. There is no indication that the vitamin D deficiency is such that it can lead to rickets.
5. Swaddled infants who are briefly separated from their mothers after birth or are placed in the nursery and receive supplements have a delayed weight-loss recovery.

As soon as traditional swaddling practices are introduced into a Western society with (centrally) heated houses and highly insulating bedding, one should be aware of the possible adverse effects of these practices and discuss them with these ethnic groups. In the Netherlands, 80% of Turkish and Moroccan parents were swaddled in infancy.[34] Nowadays, 15% of the Turkish children and 20% of the Moroccan children living in the Netherlands are swaddled, whereas ~6% of Dutch infants are swaddled.[44] Observation and clinical practice shows that these ethnic groups often swaddle inappropriately; the legs and hips are swaddled very tightly in extension and adduction. Special support and education is necessary for these ethnic groups. The risk for SIDS seems to be increased by sleeping in a prone position when swaddled and by swaddling with the head covered. In a supine position, swaddling seems to be protective, although this differs with the way that swaddling is applied. Up to a certain age, swaddling hinders turning prone, but on the other hand, when an infant is prone, his or her risk of SIDS significantly increases.

Conclusions

There are many different insights into the technique of swaddling. It has been used by many cultures and in many ways. When not used properly, swaddling can be a dangerous intervention that increases the risk of DDH, SIDS, and overheating. It can be used safely, however, if accompanied by advice about sleeping position, the way the child is swaddled (head free), the amount of (extra) bedding, and the tightness of the swaddling around the chest, hips, and knees. Preterm infants can benefit from swaddling as well, although the technique might need to differ from postnatal swaddling of full-grown infants. The issue that needs replication, however, is whether other methods of infant restraint that carry hardly any risk, such as firm tucking-in, can have the same effect on crying and sleep behaviour.

References

1. Lipton EL, Steinschneider A, Richmond JB. Swaddling, a child care practice: historical, cultural and experimental observations. Pediatrics 1965;35(suppl):521–67
2. Karp H. The Happiest Baby on the Block. London, United Kingdom: Penguin Group; 2002
3. Blom MA. Crying and Restlessness in Babies: A Parent's Guide to Natural Sleeping. Edinburgh, Scotland: Floris; 2005
4. Masataka N. On the function of swaddling as traditional infant care practiced by native South Americans [in Japanese]. Shinrigaku Kenkyu 1996;67:285–91
5. Luke 2:7 (NIV)
6. Chisholm JS. Swaddling, cradleboards and the development of children. Early Hum Dev 1978;2:255–75
7. Barry H, Paxson LM. Infancy and early childhood: crosscultural codes 2. Ethnology 1971;10:466–508
8. Kremli MK, Alshahid AH, Khoshhal KI, Zanzam MM. The pattern of developmental dysplasia of the hip. Saudi Med J 2003;24:1118–20
9. Barr RG, Konner M, Bakeman R, Adamson L. Crying in !Kung San infants: a test of the cultural specificity hypothesis. Dev Med Child Neurol 1991;33:601–10
10. Rousseau JJ. Emile. Foxley B, trans-ed. London, United Kingdom: Everyman's Library, Dent; 1911
11. Gorer G. Some aspects of the psychology of the people of Great Russia. Am Slav East Eur Rev 1949;8:155–160
12. Gorer G, Rickman J. The People of Great Russia: A Psychological Study. London, New York, NY: Cresset; 1949
13. Benedict R. Child rearing practices in certain European countries. Am J Orthopsychiatry 1949;19:342–50
14. Mead M. The swaddling hypothesis: its reception. Am Anthropol 1954;56:395–409
15. Kutlu A, Memik R, Mutlu M, Kutlu R, Arslan A. Congenital dislocation of the hip and its relation to swaddling used in Turkey. J Pediatr Orthop 1992;12:598–602
16. Yurdakok K, Yavuz T, Taylor CE. Swaddling and acute respiratory infections. Am J Public Health 1990;80:873–5
17. Wilson CA, Taylor BJ, Laing RM, Williams SM, Mitchell EA. Clothing and bedding and its relevance to sudden infant death syndrome. J Paediatr Child Health 1994;30:506–12
18. van Schaijk M, Lanting CI, van Wouwe JP, Engelberts AC, L'Hoir MP. Survey risk factors for cot death, 2002–2003 [in Dutch]. In: Van Schaijk M, Lanting CI, van Wouwe JP, Engelberts AC, L'Hoir MP, eds. TNO: Quality of Life. Leiden, Netherlands; 2006

19. Weston J. The pathology of child abuse. In: Helfer R, Kempe C, eds. The Battered Child. Chicago, IL: University of Chicago Press; 1968
20. Gerard CM, Harris KA, Thach BT. Spontaneous arousals in supine infants while swaddled and unswaddled during rapid eye movement and quiet sleep. Pediatrics 2002;110:e70
21. Short MA, Brooks-Brunn JA, Reeves DS, Yeager J, Thorpe JA. The effects of swaddling versus standard positioning on neuromuscular development in very low birth weight infants. Neonatal Netw 1996;15:25–31
22. Neu M, Browne JV. Infant physiologic and behavioral organization during swaddled versus unswaddled weighing. J Perinatol 1997;17:193–8
23. Ohgi S, Akiyama T, Arisawa K, Shigermori K. Randomised controlled trial of swaddling versus massage in the management of excessive crying in infants with cerebral injuries. Arch Dis Child 2004;89:212–16
24. Campos RG. Soothing pain-elicted distress in infants with swaddling and pacifiers. Child Dev. 1989;60:781–92
25. Short MA. A comparison of temperature in VLBW infants swaddled versus unswaddled in a double-wall incubator in skin control mode. Neonatal Netw 1998;17:25–31
26. Huang CM, Tung WS, Kuo LL, Ying-Ju C. Comparison of pain responses of premature infants to the heelstick between containment and swaddling. J Nurs Res 2004;12:31–40
27. Tsogt B, Maniseki-Holland S, Pollock J, Blair P, Fleming P. Can traditional care influence thermoregulation? A prospective controlled study of the effects of swaddling on infants' thermal balance in a Mongolian winter. Presented at: 9th SIDS Conference; June 1–4, 2006; Yokohama, Japan
28. Bystrova K, Matthiesen AS, Widström AM, et al. The effect of Russian maternity home routines on breastfeeding and neonatal weight loss with special reference to swaddling. Early Hum Dev 2007;83:29–39
29. Çağlayan S, Yaprak I, Seçkin E, Kansoy S, Aydinlioğlu H. A different approach to sleep problems of infancy: swaddling above the waist. Turk J Pediatr 1991;33:117–20
30. Chisholm JS. Navajo Infancy: An Ethological Study of Child Development. New York, NY: Aldline; 1983
31. Franco P, Seret N, van Hees JN, Scaillet S, Groswasser J, Kahn A. Influence of swaddling on sleep and arousal characteristics of healthy infants. Pediatrics 2005;115:1307–11
32. Franco P, Scaillet S, Groswasser J, Kahn A. Increased cardiac autonomic responses to auditory challenges in swaddled infants. Sleep 2004;27:1527–32
33. Kahn A, Rebuffat E, Sottiaux M. Effects of body movement restraint on cardiac response to auditory stimulation in sleeping infants. Acta Paediatr 1992;81:959–61
34. Gerard CM, Harris KA, Thach BT. Physiologic studies on swaddling: an ancient child care practice, which may promote the supine position for infant sleep. J Pediatr 2002;141:398–404

35. Giacoman SL. Hunger and motor restraint on arousal and visual attention in the infant. Child Dev 1971;42:605–14
36. Li Y, Liu J, Liu F, Guo G, Anme T, Ushijima H. Maternal child-rearing behaviors and correlates in rural minority areas of Yunnan, China. J Dev Behav Pediatr 2000;21:114–22
37. Tronick EZ, Thomas RB, Daltabuit M. The Quechua Manta Pouch: a caretaking practice for buffering the Peruvian infant against the multiple stressors of high altitude. Child Dev 1994;65:1005–13
38. Manaseki S. Mongolia: a health system in transition. BMJ 1993;307:1609–11
39. Dennis N. The Hopi Child. New York, NY: Science Editions, John Wiley; 1940
40. Leighton D, Kluckhohn C. Children of the People. Cambridge, MA: Harvard University Press; 1947
41. Dennis W, Dennis MG. The effect of cradling practices upon the onset of walking in Hopi children. J Genet Psychol 1940;56:77–86
42. Harriman AE, Lukosius PA. On why Wayne Dennis found Hopi infants retarded in age at onset of walking. Percept Mot Skills 1982;55:79–86
43. Hudson C. Isometric advantages of the cradle board. Am Anthropol 1966;68:470–4
44. van Sleuwen BE, L'Hoir MP, Engelberts AC, Westers P, Schulpen TWJ. Infant care practices related to cot death in Turkish and Moroccan families in the Netherlands. Arch Dis Child 2003;88:784–8
45. Ponsonby AL, Dwyer T, Gibbons LE, Cochrane JA, Wang YG. Factors potentiating the risk of sudden infant death syndrome associated with the prone position. N Engl J Med 1993;329:377–82
46. Beal S, Porter C. Sudden infant death syndrome related to climate. Acta Paediatr Scand 1991;80:278–7
47. Nelson EAS, Schiefenhoevel W, Haimerl F. Child care practices in nonindustrialized societies. Pediatrics 2000;105:e75
48. Jobe AH. The editors' perspectives: swaddling to promote sleep and decrease SIDS in infants [abstract]. J Pediatr 2002;141:1A
49. L'Hoir MP, Engelberts AC, van Well GTJ, et al. Risk and preventive factors for cot death in the Netherlands, a lowincidence country. Eur J Pediatr 1998;157:681–8
50. Beltman M. Swaddling of Infants: An Old Fashioned Habit on Its Return [MSc degree paper]. Utrecht, Netherlands: Utrecht University; 2000
51. Tonkin S, Hassall I. Infant sleeping position and cot death. Aust Paediatr J 1989;25:376–7
52. Chapell MS. Sudden infant death syndrome (crib death) and possible relation to long-term mattress compression. Psychol Rep 1990;67:1267–72
53. Wilson CE. Cree infant care practices and sudden infant death syndrome. Can J Public Health 2000;91:133–6
54. van Gestel JPJ, L'Hoir MP, ten Berge M, Jansen NJG, Plötz FB. Risks of ancient practices in modern times. Pediatrics 2002;110:e78

55. Bolton DP, Nelson EA, Taylor BJ, Weatherall IL. Thermal balance in infants. J Appl Physiol 1996;80:2234–42
56. Cheng TL, Partridge JC. Effect of bundling and high environmental temperature on neonatal body temperature. Pediatrics 1993;92:238–40
57. Bacon CJ, Bell SA, Clulow EE, Beattie AB. How mothers keep their babies warm. Arch Dis Child 1991;66:627–32
58. Williams SM, Taylor BJ, Mitchell EA. Sudden infant death syndrome: insulation from bedding and clothing and its effect modifiers. The National Cot Death Study Group. Int J Epidemiol 1996;25:366–75
59. Wayse V, Yousafzai A, Mogale K, Filteau S. Association of subclinical vitamin D deficiency with severe acute lower respiratory infection in Indian children under 5 y. Eur J Clin Nutr 2004;58:563–7
60. Li Z, He G, Rong H, Wu S. Effects of wrapped supine, unwrapped supine and prone positions on the transcutaneous oxygen tension in neonatal pneumonia. Chin Med J 1986;99:945–7
61. Chaisupamongkollarp T, Preuthipan A, Vaicheeta S, Chantarojanasiri T, Kongvivekkajornkij W, Suwanjutha S. Prone position in spontaneously breathing infants with pneumonia. Acta Paediatr 1999;88:1033–4
62. Urnaa V, Kizuki M, Nakamura K, et al. Association of swaddling, rickets onset and bone properties in children in Ulaanbaatar, Mongolia. Public Health 2006;120:834–40
63. Klisic PJ. Congenital dislocation of the hip: a misleading term—brief report. J Bone Joint Surg Br 1989;71:136
64. Salter RB. Etiology, pathogenesis and possible prevention of congenital dislocation of the hip. Can Med Assoc J 1968;98:933–45
65. Yamamuro T, Ishida K. Recent advances in the prevention, early diagnosis and treatment of congenital dislocation of the hip in Japan. Clin Orthop Relat Res 1984;184:34–40
66. Rabin DL, Barnett CR, Arnold WD, Freiberger RH, Brooks G. Untreated congenital hip disease: a study of the epidemiology, natural history, and social aspects of the disease in a Navajo population. Am J Public Health Nations Health 1965;55(suppl):1–44
67. Ishida K. Prevention of the development of the typical dislocation of the hip. Clin Orthop Relat Res 1977;126:167–9
68. Ishida K. A study of newborn examination for CDH with special reference to its preventive measures [in Japanese]. Orthop Surg 1973;24:1299
69. Abd el-Kader Shaheen M. Mehad: the Saudi tradition of infant wrapping as a possible etiological factor in congenital dislocation of the hip. J R Coll Surg Edinb 1989;34:85–7
70. Wedge JH, Wasylenko MJ. The natural history of congenital dislocation of the hip: a critical review. Clin Orthop Relat Res 1978;137:154–61
71. Pratt WD, Freiberger RH, Arnold WD. Untreated congenital hip dysplasia in the Navajo. Clin Orthop Relat Res 1982;162:69–77

72. Chaarani MW, Al Mahmeid MS, Salman AM. Developmental dysplasia of the hip before and after increasing community awareness of the harmful effects of swaddling. Qatar Med J 2002;11:40–3
73. Şahin F, Aktürk A, Beyazova U, Beyazova U, Çakir B, Boyunaga O, Tezcan S, Bölükbaşi S, Kanatli U. Screening for developmental dysplasia of the hip: results of a 7-year follow-up study. Pediatr Int 2004;46:162–6
74. O'Sullivan ME, O'Brien T. Acetabular dysplasia presenting as developmental dislocation of the hip. J Pediatr Orthop 1994;14:13–5
75. von Rosen S. Diagnosis and treatment of congenital dislocation of the hip joint in the newborn. J Bone Joint Surg Br 1962;44:284
76. Salter RB, Kostuik J, Schatzker J. Experimental dysplasia of the hip and its reversibility in newborn pigs. J Bone Joint Surg 1963;45:1781
77. Fearon I, Kisilevsky BS, Hains SMJ, Muir DW, Tranmer J. Swaddling after heel lance: age-specific effects on behavioral recovery in preterm infants. J Dev Behav Pediatr 1997;18:222–32
78. Huhtala V, Lehtonen L, Heinonen R, Korvenranta H. Infant massage compared to a crib vibrator in the treatment of colicky infants. Pediatrics 2000;105:e84
79. Lehtonen L, Korvenranta H. Infantile colic: seasonal incidence and crying profiles. Arch Pediatr Adolesc Med 1995;149:533–6
80. Saylor C, Lippa B, Lee G. Drug-exposed infants at home: strategies and support. Public Health Nurs 1991;8:33–8
81. van Sleuwen BE, L'Hoir MP, Engelberts AC, Busschers WB, Westers P, Blom MA, Schulpen TWJ, Kuis W. Comparison of behavior modification with and without swaddling as interventions for excessive crying. J Pediatr 2006;149:512–7
82. Thompson L, Joseph A. Childhood the Hopi way. In: Seidman JM, ed. The Child: A Book of Readings. New York, NY: Rinehart; 1958

Infant care practices related to cot death in Turkish and Moroccan families in the Netherlands

3

Bregje E. van Sleuwen
Monique P. L'Hoir
Adèle C. Engelberts
Paul Westers
Tom W.J. Schulpen

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Abstract

From 1979 to 1993 Turkish infants had a significantly higher cot death risk compared to Dutch infants. In contrast Moroccan infants had a risk of cot death that was approximately three times lower compared to Dutch infants during the same period. This study shows that these differences have disappeared, while differences still exist in infant care practices between these ethnic groups. At 28 well-baby clinics, questionnaires were distributed for this sample selection. The response was 82%. Data were collected on 55 Turkish, 54 Moroccan, and 210 Dutch families.
Less than 7% of these three ethnic groups still placed infants in the prone position. Moroccan mothers hardly smoked. Turkish people used pillows and Moroccan people used soft mattresses more often. Moroccan families practised swaddling more widely. Length of maternal residence influenced some care giving practices. As a result of this study, subgroup specific intervention campaigns for safe sleeping can be developed for Turkish and Moroccan families.

Keywords

cot death, sudden infant death syndrome, infant care practices, ethnic group

The Netherlands with its 16 million inhabitants has a relatively large ethnic minority of 1.3 million persons, amounting to nearly 9% of the population. The largest group came to our country as migrant labourer in the 1960s from Islamic Mediterranean countries, chiefly Turkey and Morocco. The other major group consists of people from Surinam in South America and from the Netherlands Antilles in the Caribbean, both former Dutch colonies. Nearly half of the migrant population lives in the four major cities (Amsterdam, Rotterdam, Utrecht, and The Hague), where 15% of the population and 40–50% of the children are of foreign descent.

In a retrospective analysis of 20.211 death certificates of all children who died between 1979 and 1993, data about cot death were collected for different ethnic groups.[1,2] The incidence of sudden and unexpected death was 1.65 per 1000 live born Turkish infants. In contrast, the Moroccan infants had an incidence of 0.40. Compared to Dutch infants, whose cot death incidence was 1.4 per 1000 live births, the risk of cot death was significantly higher in Turkish infants and more than three times lower in Moroccan infants. There was no readily available explanation for this difference, except that Turkish fathers as well as mothers smoke far more heavily than Moroccan parents. To explore the reasons behind these incidence differences further, the current study on infant care practices was performed.

In 1999, Van der Wal and colleagues[3] examined the care practices of 1529 infants—114 Turkish infants, 225 Moroccan infants, 181 Surinam infants, 775 Dutch infants, and 234 infants of other descent. Prenatal smoking was low in Moroccan mothers (0.4%), while Turkish mothers smoked almost as much as Dutch mothers (18.5% and 21.6% respectively). Daily smoking in the home occurred more often in Turkish families, compared to the Moroccan and Dutch homes. Turkish infants were placed prone more often than Moroccan and Dutch infants, but this difference was not statistically significant. Turkish and Moroccan families did not differ concerning breastfeeding. The use of duvets and a pillow was higher among Turkish and Moroccan infants compared to Dutch infants.

Several risk and preventive factors for cot death were not covered in the study of Van der Wal and colleagues,[3] however. In order to investigate what other differences between ethnic groups exist in infants care practices, the present study was carried out. Next to the previous mentioned aspects of infant care, the study focused on overheating, ventilation, room and bed sharing, type of mattress, dummy use, and swaddling.

Methods

A pilot study was carried out, including 22 Turkish, 44 Moroccan, and 31 Dutch families living in the Utrecht area.[4] After analysis the questionnaire was adjusted on several items and prepared for the present study. From the Dutch Central Bureau of Statistics a list of villages and cities was gathered in which more than 500 Turkish and more than 500 Moroccan families lived.[5] Eight regions, including urban and suburban areas, were randomly selected. It was a representative sample of concentrations of

Turkish and Moroccan populations. Data were collected by face to face interviews from June 1999 to October 1999 by two previously trained interviewers at 28 well-baby clinics. From the pilot study we learned that many ethnic mothers who did not speak Dutch, were accompanied by a family member who did speak Dutch. For practical and financial reasons it was decided not to use interpreters. During consulting hours at well-baby clinics all parents of infants between 4 weeks and 2 years of age, not only Turkish and Moroccan and Dutch parents, were asked to participate. Ethnicity of the child was defined as the ethnicity of the mother. The final question in the questionnaire was if the parents had ever heard of cot death.
The thermal resistance of the total number of layers bedding was defined by tog values. A tog is a unit of thermal resistance, one tog being 10 times the temperature difference in degrees Celsius between the surfaces of a material when the heat flow through it is 1 watt/m^{2},[6] Tog values were estimated from the usual use of bedclothes as reported by the parents.

Statistical methods

Correlation analyses, using Spearman's rho and logistic regression analyses were used to analyse the data, using the Statistical Package for the Social Sciences (SPSS) version 8. χ^2 tests, analysis of variance, and *t* tests were used to analyse the demographic data and the tog values. Confounders were selected in advance from earlier Dutch studies, literature review, and ongoing international research. They were: age of the mother, age of the infant, birth order, and maternal education. All p values are two tailed.

Results

Data were collected on 55 Turkish, 54 Moroccan, and 210 Dutch families, and 33 families of other ethnic decent, giving a response of 83%. The 33 families of other descent were subsequently excluded in the study, as the number of families per ethnic group was too small. Forty-one parents refused participation, four infants in retrospect did not meet the age criteria, and 32 parents did not participate because of language difficulties.
The following demographic data were collected: gender, birth order, birth interval, parental education, maternal smoking at time of the interview and maternal smoking during pregnancy, age of the infant, age of parents, age of mother at first live birth, country of birth of the parents, and length of stay in the Netherlands (Table 1). The Turkish and Moroccan people did not differ in most of the demographic characteristics, except that the Moroccan infants were significantly younger than the Dutch and Turkish infants. The Dutch parents were higher educated.

We checked by multivariate analysis whether some age related factors, such as room sharing, bed sharing, use of a pillow, use of a duvet during summer and winter, and dummy use were confounded. Some of these factors were influenced by age; for those factors, age specific groups were made.

Table 1. *Demographic factors.*

	Dutch	Turkish	Moroccan
Gender*			
Boy	119 (56.7)	33 (60.0)	28 (51.9)
Girl	91 (43.3)	22 (40.0)	26 (48.1)
Birth order*			
First born	120 (57.1)	14 (25.5)	16 (29.6)
Second born	71 (3.8)	19 (34.5)	17 (31.5)
Third born or more	19 (9.0)	22 (40.0)	21 (38.9)
Maternal education*			
Primary school	11 (5.2)	28 (53.8)	16 (29.6)
Lbo/mavo@	53 (25.2)	14 (26.9)	21 (38.9)
Havo/vwo/mbo$	86 (41.0)	9 (17.3)	14 (25.9)
Hbo/university^	60 (28.6)	1 (1.9)	3 (5.6)
Paternal education*			
Primary school	6 (3.0)	16 (31.4)	15 (28.3)
Lbo/mavo@	55 (27.9)	17 (33.3)	19 (35.8)
Havo/vwo/mbo$	70 (35.5)	14 (27.5)	11 (20.8)
Hbo/university^	66 (33.5)	4 (7.8)	8 (15.1)
Maternal smoking at time of interview*	69 (32.9)	16 (29.6)	1 (1.9)
Maternal smoking during pregnancy*	56 (26.7)	12 (22.2)	1 (1.9)
Age of the infant (months)#	7.45 ± 4.83	7.42 ± 5.22	5.74 ± 3.61
Age of mother (years)#	30.38 ± 5.23	28.65 ± 5.75	27.72 ± 4.38
Age mother at first live birth (years)#	27.57 ± 4.98	21.67 ± 3.48	22.02 ± 3.27
Age of father (years)#	33.14 ± 6.09	31.25 ± 7.38	33.81 ± 5.56
Length of maternal residence in the Netherlands#	30.24 ± 5.54	11.84 ± 7.74	12.80 ± 7.51
Length of paternal residence in the Netherlands#	31.65 ± 7.91	13.01 ± 8.62	13.36 ± 7.96

* Counts (%)
\# Mean ± SD

@ Lower technical and vocational training and lower general secondary education
$ Intermediate vocational training and advanced secondary education
^ Higher vocational education (college education) and university

Sleeping position

The prevalence of prone sleeping was very low (<10%) in all groups (Table 2). Moroccan parents and Turkish parents placed infants in the side position to sleep more often, and in the supine position less often compared to Dutch parents.

Bedding

Pillows were used more often in Moroccan families than in Dutch ones, but in Turkish families they were used even more so (Table 2). Moroccan people compared to the Dutch families used soft mattresses most often. Plastic covers to protect the mattress

are used equally often by all three groups. Cot buffers were used equally often in Turkish and Moroccan families, but significantly more than in Dutch families.
Fifty three per cent of the Turkish, 35% of the Moroccan, and 71% of the Dutch group used a Dutch sleeping sack. Blanket use, however, did not differ significantly between the groups, neither during summer nor during wintertime, and is therefore not noted in the table.
In this study only 25% of the Dutch parents used duvets. However, 55% of both Turkish and Moroccan people used duvets in wintertime.

Tog values

The Turkish and Moroccan population use duvets in summertime more often than Dutch families. However, after adjustment for "birth order" and "level of education of the mother", the difference is not statistically significant. Low level of education was related to duvet use in summertime (r = 0.24, $p < 0.0005$). The estimated total amount of tog values in summer is significantly higher for Turkish (mean 4.71 (SD 3.95)) and Moroccan people (3.73 (SD 4.11)) compared to Dutch people (mean 2.55 (SD 3.03); $p < 0.0005$). In wintertime the statistically significant difference in tog values between these three groups remains ($p = 0.005$).

Central heating

In summer and in wintertime, Turkish and Moroccan families have the central heating on more often compared to Dutch families (Table 3). The room temperature in one fourth of Turkish and Moroccan families is higher than 20 degrees, while this is the case in 17% of the Dutch families. These differences are not statistically significant, however.

Keeping a window open in the infant's room seems to be culturally related; one third of the Dutch parents, less than one fourth of the Turkish, and one sixth of the Moroccan families have a window open. It was also asked whether the parents ventilated the infant's room. Almost all parents of all ethnic groups either did ventilate the room or had a door or window open in the infant's room.

Table 2. Relation between ethnicity and demographic factors* on sleeping position and bedding.

	Ethnicity Dutch	Turkish			Moroccan		
Infant care practice	N (%)	N (%)	OR (95% CI) Univariate	OR (95% CI) Multivariate	N (%)	OR (95% CI) Univariate	OR (95% CI) Multivariate
Sleeping position							
Supine	176 (84.6)	37 (74.0)	0.52 (0.25 to 1.08)	0.55 (0.25 to 1.19)	34 (69.4)	0.41 (0.20 to 0.84)	0.34 (0.16 to 0.71)
Prone	11 (5.3)	3 (6.0)	1.14 (0.31 to 4.26)	1.10 (0.29 to 4.23)	2 (4.1)	0.76 (0.16 to 3.55)	0.97 (0.20 to 1.55)
Side	21 (10.1)	10 (20.0)	2.22 (0.97 to 5.09)	2.11 (0.90 to 4.96)	13 (26.5)	3.21 (1.48 to 7.00)	3.21 (1.48 to 7.00)
Bedding							
Duvet use during summer							
0-6 months	11 (11.8)	12 (48.0)	6.90 (2.52 to 18.81)	3.42 (1.01 to 11.69)	10 (33.3)	3.73 (1.39 to 9.99)	1.69 (0.54 to 5.32)
6-12 months	13 (16.9)	5 (35.7)	2.73 (0.78 to 0.49)	2.73 (0.79 to 9.49)	5 (23.8)	1.54 (0.48 to 4.95)	1.54 (0.48 to 4.95)
> 12 months	5 (12.8)	4 (26.7)	2.48 (0.56 to 10.86)	1.86 (0.38 to 9.04)	1 (50.0)	6.80 (0.36 to 126.88)	6.80 (0.36 to 126.88)
Duvet use during winter							
0-6 months	20 (22.2)	14 (56.0)	4.46 (1.75 to 11.32)	1.94 (0.62 to 6.12)	16 (51.6)	3.73 (1.58 to 8.84)	1.72 (0.64 to 4.61)
6-12 months	17 (22.1)	9 (64.3)	6.37 (1.88 to 21.50)	3.92 (0.79 to 19.5)	12 (57.1)	4.69 (1.70 to 13.04)	2.54 (0.73 to 8.85)
> 12 months	14 (35.9)	7 (46.7)	1.56 (0.47 to 5.22)	1.34 (0.39 to 4.65)	2 (100.0)	-	-
Sleeping sack	148 (70.5)	29 (52.7)	0.47 (0.25 to 0.85)	0.44 (0.22 to 0.86)	19 (35.2)	0.23 (0.12 to 0.43)	0.25 (0.13 to 0.50)
Pillow	5 (2.5)	17 (31.5)	18.18 (6.32 to 52.34)	12.82 (3.07 to 53.48)	6 (11.1)	4.95 (1.45 to 16.89)	4.52 (1.04 to 19.79)
Cot buffer	23 (12.6)	21 (45.7)	5.84 (2.83 to 12.09)	5.05 (2.37 to 10.75)	20 (46.5)	6.05 (2.88 to 12.70)	5.29 (2.49 to 11.22)
Soft mattress	12 (5.7)	9 (16.4)	3.22 (1.28 to 8.11)	2.56 (0.96 to 6.89)	17 (31.5)	7.58 (3.4 to 17.16)	7.58 (3.34 to 17.16)
Plastic mattress cover	37 (17.6)	7 (12.7)	0.68 (0.29 to 1.63)	0.64 (0.26 to 1.57)	6 (11.1)	0.58 (0.23 to 1.47)	0.49 (0.19 to 1.25)

* OR's are adjusted for maternal age, age of the infant, and maternal education in the multivariate analysis

Table 3. Relation between demographic factors* and ethnicity on parental room sharing, bed sharing, sleeping separately, and swaddling.

	Ethnicity Dutch	Turkish			Moroccan		
Infant care practice	N (%)	N (%)	OR (95% CI) Univariate	OR (95% CI) Multivariate	N (%)	OR (95% CI) Univariate	OR (95% CI) Multivariate
Central heating							
In summer	11 (0.5)	7 (13.0)	31.15 (3.74 to 259.06)	27.86 (3.27 to 237.15)	3 (5.6)	12.30 (1.25 to 120.65)	12.30 (1.25 to 120.65)
In winter	66 (31.7)	48 (87.3)	14.73 (6.33 to 34.30)	3.81 (5.92 to 32.27)	44 (81.5)	9.47 (4.49 to 19.96)	9.47 (4.49 to 19.96)
Parental room sharing (without bed sharing), bed sharing, and sleeping separately							
Parental room sharing							
0-2 months	12 (42.9)	2 (22.2)	0.38 (0.07 to 2.17)	0.19 (0.02 to 1.76)	6 (54.5)	1.60 (0.39 to 6.51)	1.60 (0.39 to 6.51)
2-3 months	3 (17.6)	1 (25.0)	1.56 (0.12 to 20.60)	1.56 (0.12 to 20.60)	0 (0)	-	-
> 3 months	12 (7.3)	11 (26.8)	4.67 (1.89 to 11.58)	3.82 (1.48 to 9.87)	11 (26.2)	4.52 (1.83 to 11.18)	4.52 (1.83 to 11.18)
Bed sharing (occasionally)	84 (40.4)	20 (37.0)	0.87 (0.47 to 1.61)	0.85 (0.45 to 1.59)	21 (38.9)	0.94 (0.51 to 1.72)	0.81 (0.43 to 1.51)
Door of room for baby open (no bed sharing nor room sharing)	56 (47.5)	11 (45.8)	0.93 (0.39 to 2.27)	0.93 (0.39 to 2.27)	17 (73.9)	3.13 (1.15 to 8.33)	3.13 (1.15 to 8.33)
Dummy use							
0-3 months	18 (40.0)	10 (76.9)	5.00 (1.21 to 20.71)	7.50 (1.47 to 38.31)	6 (50.0)	1.50 (0.42 to 5.39)	1.50 (0.42 to 5.39)
3-6 months	17 (34.7)	7 (58.3)	2.64 (0.73 to 9.57)	2.26 (0.60 to 8.49)	13 (68.4)	4.08 (1.31 to 12.65)	4.08 (0.27 to 12.65)
> 6 months	54 (47.4)	11 (39.3)	0.72 (0.31 to 1.67)	0.76 (0.33 to 1.79)	13 (59.1)	1.61 (0.64 to 4.05)	1.61 (0.64 to 4.05)
Swaddling							
Swaddling infant	13 (6.2)	6 (10.9)	1.85 (0.67 to 5.00)	1.65 (0.56 to 4.85)	8 (14.8)	2.63 (1.03 to 6.67)	2.63 (1.03 to 6.73)
Swaddling mother in infancy	13 (6.7)	44 (86.3)	100 (33.33 to 10000)	100 (34.33 to 266.69)	9 (75.0)	50.00 (20.00 to 100)	41.67 (18.07 to 97.49)

* OR's are adjusted for maternal age, age of the infant, and maternal education in the multivariate analysis

Bed sharing, room sharing, and leaving the door open

Room sharing (excluding bed sharing) occurred in one third of the Moroccan families (Table 3). When the child sleeps in a separate room, Moroccan families often keep the door of that room open (OR 3.13, 95% CI 1.15 to 8.33), compared to Dutch families.

Maternal smoking

Moroccan mothers smoked significant less during pregnancy and at the time of the interview than Turkish and Dutch mothers (χ^2, $p = 0.01$ resp. χ^2, $p = 0.006$).

Dummy use, thumb/finger sucking, and mouth breathing

Moroccan infants were offered a dummy most often. In the multivariate analysis, however, after introducing the factor "birth order", the difference loses its statistical significance. Moroccan infants appear to breathe through their mouth* and suck their thumb/finger less often than Turkish and Dutch infants. The mouth breathing effect disappeared in the multivariate analysis but thumb/finger sucking remained statistically significant ($p < 0.01$). Thumb sucking occurs in 50.5% of the Dutch, in 40.0% of the Turkish, and 35.2% of the Moroccan infants. The correlation between dummy use and thumb/finger sucking is statistically significant ($r = -0.12$, $p = 0.03$) as well as between thumb/finger sucking and mouth breathing ($r = 0.13$, $p = 0.03$), although it is not very strong.

Swaddling

Moroccan infants were swaddled more often compared to Dutch and Turkish infants (OR 2.63, 95% CI 1.03 to 6.73) (Table 3). In the pilot study we had already found that Moroccan people swaddled their infants more often (29.5%) than Dutch (0%) and Turkish (13.7%) families. In the current study the mothers themselves had been swaddled more often in the ethnic groups than the Dutch mothers had been (Turkish 86.3%, Moroccan 75%, and Dutch 6.7%).

Information about prevention of cot death

Half of the Turkish and Moroccan families did not know anything about cot death. Forty five per cent of the Turkish people and 52% of the Moroccans had never heard of it before. Only 3% of the Dutch people had never heard of cot death before.

Length of maternal residence in the Netherlands

A longer period of stay in the Netherlands is significantly ($p < 0.05$) correlated with less use of a pillow ($r = 0.25$), less use of a cot buffer ($r = 0.26$), and less use of central heating ($r = 0.21$). Length of maternal stay had no effect on smoking behaviour of the mothers.

Discussion

A large study of infant mortality in immigrant groups, covering the period from 1979 to 1993, showed an increased risk of cot death for Turks, but a decreased risk for Moroccans compared to Dutch infants.[1] However, the increased cot death incidence of the Turkish decreased proportionally with the Dutch decrease in incidence.[8,9] All in all, cot death differences between the Turkish and Moroccan populations have almost disappeared (1996–2000, Turkish 0.24, Moroccan 0.28, and Dutch 0.16 per 1000 live born infants). This study looks at cultural differences in infant care practices related to cot death in the light of a historical difference in incidence between three ethnic populations. Understanding such differences is important in furthering knowledge of how infant care practice may relate to cot death.

A limitation of this study is a response of 83%, and the lack of data on the non-response group.

Summarising, both Turkish and Moroccan mothers were less likely to lay their baby supine, less likely to use a sleeping sack, and more likely to use a pillow or cot buffer. Turkish mothers in particular were more likely to use a pillow, while Moroccan mothers were more likely to have a soft mattress. Both had higher tog values both in the summer and winter. Conversely there were protective factors in that both groups were more likely to share the parental room without bed sharing, less likely to smoke (especially Moroccan mothers), and more likely to swaddle.

A full explanation for the rather constant incidence of cot death in Moroccan infants cannot be given. Moroccan people have several customs which are known to be associated with a decreased risk of cot death, such as little maternal smoking and room sharing, and perhaps factors such as swaddling and leaving the bedroom door open, thus improving parental surveillance. Since we know that half of the Dutch children that are still being placed prone (8%) are children that cry excessively,[9,10] we speculated that in the Moroccan population the prone sleeping group could be underrepresented, because they swaddle these infants.[11]

Finally, it has been described that people from Southern European countries promote dummy use as a measure to prevent thumb sucking. Mouth breathing occurs less in these countries.[7,12] It has been established in many studies that dummy use, especially during the last sleep, lowers the risk of cot death.[12–16] It has been reported that people from Southern European countries (Turkey, Spain, and Morocco) have a low prevalence of thumb sucking because mothers consciously prevent this by giving the breast or a dummy.[7] The social stigma against thumb sucking in the Southern European cultures thus appears potentially protective against cot death.

However, since most risk and protective factors are shared by both Turkish and Moroccan families, cultural infant care practices cannot really account for the observed differences.

Information on cot death does not seem to reach this group of young ethnic families. Many parents did not even know of the concept of cot death, let alone of important risk factors.

Differences that exist in infant care practices between Turkish and Moroccan populations have their origin in old habits and customs that by chance seem to be either preventive or risk increasing. It is very important for the prevention of cot death to keep in mind the ethnic differences in childcare practices. It is important to inform ethnic minorities about their native protective customs such as non-prone sleeping and perhaps swaddling which was recently postulated as being protective,[17] and to warn them against a preference for side sleeping and pillows, and against several Western habits, such as the use of a duvet. The length of maternal residence did influence several risk and preventive factors positively, which suggests that the Dutch prevention campaigns reached these parents. Special attention in health education of ethnic groups should be given to overheating of the child, smoking, the use of a duvet, the number of layers used in bedding, and the use of a pillow. As a result of this study language specific intervention campaigns for safe sleeping can be developed for Turkish and Moroccan families.

Footnotes

* "Sleeping with an open mouth" is termed "mouth breathing"; however, only 50% of infants who sleep with an open mouth are in fact "mouth breathing"; the other 50% still breathe through the nose despite an open mouth, by pressing the tongue to the palate.[7]

References

1. Schulpen TWJ, Steenbergen JE van, Driel HF van. Influences of ethnicity on perinatal and child mortality in the Netherlands. Arch Dis Child 2001;84:222–6
2. Dutch Central Bureau of Statistics (CBS). Mortality, cause of death, age and gender in 1969–1999 [in Dutch]. Voorburg, 2000
3. Wal MF van der, Jonge GA de, Pauw-Plomp H. Ethnic origin and care giving styles relevant to cot death [in Dutch]. Ned Tijdschr Geneesk 1999;43:143
4. Hamidzai M, L'Hoir MP, Schulpen TWJ. Cot death and differences in infant care practices of Turkish and Moroccan families, a pilot study [in Dutch]. Utrecht: Centre for Migration and Child Health, Wilhelmina Children's Hospital, 1998
5. Dutch Central Bureau of Statistics (CBS). Non-Dutch inhabitants in the Netherlands 1998 [in Dutch]. Heerlen: CBS, 1999
6. Bacon JC, Bell SA, Clulow EE, et al. How mothers keep their babies warm. Arch Dis Child 1991;66:627–32
7. Idema NK, Damsté PH, Habitual mouthbreathing. An exploration [in Dutch]. Houten/Zaventem: Bohn Stafleu Van Loghum, 1994
8. L'Hoir MP. Cot death. Risk factors and prevention in the Netherlands in 1995–1996. Thesis, University of Utrecht, 1998
9. Jonge GA de, L'Hoir MP, Ruys JH, et al. Cot death. Experiences and insights [in Dutch]. Noorden: Dutch Cot Death Foundation, 2002
10. Wal MF van der, Boom DC van den, Pauw-Plomp H, et al. Mother's reports of crying and soothing in a multicultural population. Arch Dis Child 1998;79:312–17
11. American Academy of Paediatrics, Committee on Paediatric Research. Race/ethnicity, gender, socio-economic status-Research exploring their effect on child health: a subject review. Pediatrics 2000;105:1349–51
12. L'Hoir MP, Engelberts AC, Well GT van, et al. Dummy use, thumb sucking, mouth breathing and cot death. Eur J Pediatr 1999;158:896–901
13. Mitchell EA, Taylor BJ, Ford RP, et al. Dummies and the sudden infant death syndrome. Arch Dis Child 1993;68:501–4
14. Fleming PJ, Blair PS, Pollard K, et al. Pacifier use and sudden infant death syndrome: results from the CESDI/SUDI case control study. Arch Dis Child 1999;81:112–16
15. Arnestad M, Andersen M, Rognum TO. Is the use of dummy or carry-cot of importance for sudden infant death? Eur J Pediatr 1997;156:968–70
16. Hauck FR, Herman SM, Donovan M, et al. Sleep environment and the risk of sudden infant death syndrome in an urban population: The Chicago Infant Mortality Study. Paediatrics 2003;111:1207–14
17. Gerard CM, Harris KA, Thach BT. Physiologic studies on swaddling: an ancient childcare practice, which may promote the supine position for infant sleep. J Pediatr 2002;141:398–404

Comparison of behavior modification with and without swaddling as interventions for excessive crying

4

Bregje E. van Sleuwen
Monique P. L'Hoir
Adèle C. Engelberts
Wim B. Busschers
Paul Westers
Maria A. Blom
Tom W.J. Schulpen
Wietse Kuis

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Abstract

Objective

To test the hypothesis that swaddling is an effective method to reduce crying, we compared a standardized approach of regularity and stimulus reduction with the same approach supplemented with swaddling.

Study design

Healthcare nurses coached 398 excessively crying infants up to 12 weeks of age for 3 months. Outcome measurements were crying as measured by Barr's 24-hour diary and parental perception of crying.

Results

Crying decreased by 42% in both groups after the first intervention week. Swaddling had no added benefit in the total group. Young infants (1-7 weeks of age at randomisation) benefited significantly more from swaddling as shown by a larger decrease of crying over the total intervention period. Older infants (8-13 weeks of age at randomisation) showed a significantly greater decrease in crying when offered the standardized approach without swaddling. The actual difference in crying time was 10 minutes.

Conclusion

For older babies, swaddling did not bring any benefit when added to regularity and stimuli reduction in baby care, although swaddling was a beneficial supplementation in excessively crying infants <8 weeks of age.

Excessive crying, defined as crying for more than 3 hours a day for at least one day in the preceding week, occurred in 13% and 8.6% respectively in two large Dutch prevalence studies in 1999[1] and 2003 (unpublished data). Wessel et al's definition[2] of excessive crying, namely more than 3 hours on at least 3 days of the 3 previous weeks, occurred in 2% and 2.5% of the cases. In the Netherlands 22% of parents consult a well-baby clinic doctor for excessive crying.[1] Excessive crying elicits risky behaviour in caregivers: In the Netherlands half of the 7% of parents who place their infant prone to sleep do so because their infant cries excessively or sleeps restlessly. Other consequences of excessive crying may be breast-feeding failure, attachment problems, marital stress, and postpartum depression.[3] In a recent Dutch study, 5.6% of parents of infants 6 months of age reported having smothered, slapped, or shaken their baby at least once because of its crying.[4] In 80% of the cases of child battering, excessive crying precedes the beating and inconsolable crying is a trigger event for shaking.[5,6]

Most parents, as a reaction to the crying of their infant, start carrying it, thus adding stimuli. In a randomised controlled trial such carrying has been shown to be ineffective.[7]

Instead of placing the tired infant in bed while awake, the infant falls asleep in the arms of the parent. Thus parents get more and more entangled in the sleeping practices of their child, while the child does not learn to fall asleep by itself.[8] Through lack of evidence based interventions, primary healthcare workers offer various coping strategies to parents.

Swaddling has rapidly become perceived as effective, not only by parents but also by primary healthcare workers. The introduction of yet another new intervention without scientific proof of efficacy led to this study.

We compared two interventions: behavioural modification of baby care supporting regularity and stimuli reduction, which has been shown to be effective in one controlled study,[9] and the same behavioural modification supplemented with swaddling during all sleeping periods.[8]

Methods

Design

Targeted for ascertainment were parents of 400 excessively crying infants. The intervention considered as potentially effective, namely advice concerning stimuli reduction and regularity in infant care, was offered to 200 parent-infant couples (regularity, R), whereas 200 parent-infant couples received the same advice, but their infant was also swaddled during all sleeping periods (regularity and swaddling, RS).[8-10]

A standardized case definition with the most commonly used modified Wessel et al's criteria of excessive crying (i.e., crying for more than 3 hours/24 hours for at least 3 days a week) was used for recruitment.[2] The perception of the parents and/or

doctors about the amount of crying was decisive for participation. The actual frequency and duration of crying was established later in a 24-hour diary.[11] In a baseline week, all infants received a hypoallergenic diet, either hypoallergenic formula or a hypoallergenic diet for breast-feeding mothers, to exclude infants with a possible cow's milk allergy. After this baseline week, blinded randomisation by telephone through an independent computerized centre divided the participants into one of the two groups; all parents received instruction about regularity and stimuli reduction and half of the parents were also taught to swaddle their infants during sleep.
Stratification occurred on (1) area (Utrecht, The Hague, Arnhem, and Raalte/Zwolle); (2) age (older or younger than 7 weeks); and (3) amount of crying as assessed by the parents (more or less than 5 hours/24 hours). Before randomisation, the parents were asked for their intervention preference to assess the influence of preference on outcome. The Medical Ethical Committee of the University Medical Centre Utrecht approved the study. All parents gave informed consent.

Selection

Infants were referred from February 2001 to March 2003 by well-baby clinics, general practitioners, children's hospitals, or self-referral. Inclusion criteria were healthy infants with a maximum age of 12 weeks and 6 days and a minimal gestational age of 32 weeks who cried excessively according to their parents, doctor, or healthcare worker. Children with a risk for developmental dysplasia of the hip by familial predisposition or breech position during the last month of pregnancy were excluded. Attending physicians of all participating infants filled out a form declaring normal hip function at physical examination.

Intervention

All infant-parent couples visited a specially trained healthcare nurse at one of the four locations. The consultation process, frequencies, and content as well as the method of swaddling used in this research are described by Blom et al.8 Parents were given a written outline of the study and filled out a questionnaire. The nurse instructed the parents to use a 24-hour diary for 7 days, starting that day.[11] All infants were put on a hypoallergenic diet (hypoallergenic formula or, in case of breast-feeding, a maternal diet free of milk, egg, wheat, and nut products). The project manager had a 15-minute supervision call at day 3 to verify the proper use of the 24-hour diary and the hypoallergenic diet and to check that all concomitant interventions had been stopped. After the baseline week the parents made their second visit. If the crying had been reduced to <2 hours/24 hours from day 3/4 of the baseline week onward and/or the parents perceived no more excessive crying, the infants were excluded. For all other infants the hypoallergenic diet was stopped, and the infant was randomly assigned to start of intervention (t_0).[3]

The behavioural intervention applied to both groups is based on earlier work,[8-10] and consists of a recurrent pattern of baby care, which provides structure and regularity without being rigid or inflexible. Parents applied a sequence of (1) sleeping; (2) feeding; (3) positive interaction with baby; and (4) awake time of baby alone in a playpen. Parents were instructed to watch for signs of tiredness of the baby while in the playpen. When tired, the baby was put to bed and tucked in tightly with sheet and blankets, after which a new cycle started. Essential is the repetitiveness of the elements, feeding the infant directly after waking (with an assumption that a well-rested baby is able to drink effectively) and not feeding to stop crying, and after playing alone putting the child to bed sleepy but awake. The detailed instructions were also given to parents in writing. The RS-group was given identical instructions but additionally was taught to swaddle their infant during each sleeping period. The RS-group was given the same written instructions augmented with details on how and when to swaddle the child.[8] The infant was swaddled by wrapping it in two rectangular cloths. Shoulders and arms are wrapped tightly; from the hip down the wrapping was less tight, allowing for leg flexion and abduction. Parents had to continue with the 24-hour diary on t0 for a second run of 7 days.[11] On day 2 and 5 after randomisation the nurse made a 15-minute supportive call. One week after randomisation the parents had their third visit. In a 30-minute appointment the nurse asked about the progress the parents had made and answered questions. After the first intervention week parents filled out the 24-hour diary on one fixed day a week. The project manager had a 30-minute supervision-call at week 5 and week 13 after inclusion, and the nurse had a 30-minute supportive call at week 9. In the call at week 5, in case of lack of effect perceived by the project manager and the parents, parents were offered the possibility to switch to the other intervention group. Before this supervision call parents were not informed about the possibility to change intervention.

Measurements Obtained during the First Visit

Parents recorded infant behaviour continuously in a 24- hour diary.[11] Maternal perception of infant crying and quality of the cry were recorded three times during the 12 weeks of the trial in the Cry Perception Scale[12]; twice during the baseline week, to assess reliability, and once at week 12. Furthermore, medical and psychological measures were obtained to assess their influence, as their effect on infant crying needs further explanation.[13-19]

Statistical Analysis

All data were entered and analysed in the Statistical Package for the Social Sciences for Windows 12.0 (SPSS Inc., Chicago, Ill.), Statistical Package of Social Science 2.0 (The R Foundation for Statistical Computing, Vienna University of Technology, Vienna, Austria), and MIXOR 2.0 (Discerning Systems Inc., Burnaby, Canada). T-tests, Pearson's χ^2 tests, repeated measures analyses, Mann-Whitney *U* tests, and

Analysis of Variance were used to analyse the demographic data and the Cry Perception Scale. Linear mixed models were used to account for dependencies within subjects in time, where response is adjusted by the covariate baseline. In case of non-normal distributed errors, logarithmic transformation was applied to achieve better fit. The analysis was split: We first investigated whether the odds of crying was different for the treatments using the binary logistic model for longitudinal data, and then, given that a child cried, the amount of crying was modelled in time (MIXOR 2.0, Discerning Systems Inc., Burnaby, Canada). Following the principle of intention to treat, the children who changed treatment after week 5 were analysed in the group to which they were assigned at t0.

Cry Features

From the diary we calculated the total and mean duration of crying, fussing, sleeping, being awake alone and content, playing with the baby, feeding, caring and crying/fussing (minutes/24 hours [SD]). Excessive crying was defined as crying >180 minutes/24 hours in the baseline week. In a linear mixed-effects model, the differences in the group slopes of the two curves of crying, fussing, and crying/fussing were analysed. For the baseline as well as the first week of intervention the mean of the daily diaries was used; for the long-term effect the data from the fixed day of the week were used. At first, the effect of age (groups), intervention (groups), and time on crying were analysed. These analyses were repeated with covariates.

Cry Perception Features

With repeated measurement analyses differences in scores between the baseline week and week 12 as well as differences in scores of the swaddling and regularity group were obtained from the Cry Perception Scale.[12]

Results

Response

In the study 504 infants were ascertained of whom 496 were included (Figure 1). After the baseline week with the hypoallergenic diet 47 infants (9.5%) were excluded because the parents or nurse observed that the crying was not perceived as excessive or respectively was <2 hours/24 hours from day 3/4 of the baseline week onward (average crying at day 3-7: 89.48 minutes/24 hours).
Fifty-one parents (10.3%) decided to stop participation. Finally, 398 infants (80.2%) were included. After randomisation another 16 parents (4%) quit and after 4 intervention weeks two parents in the swaddled group stopped swaddling and 16 in the regularity group started swaddling.

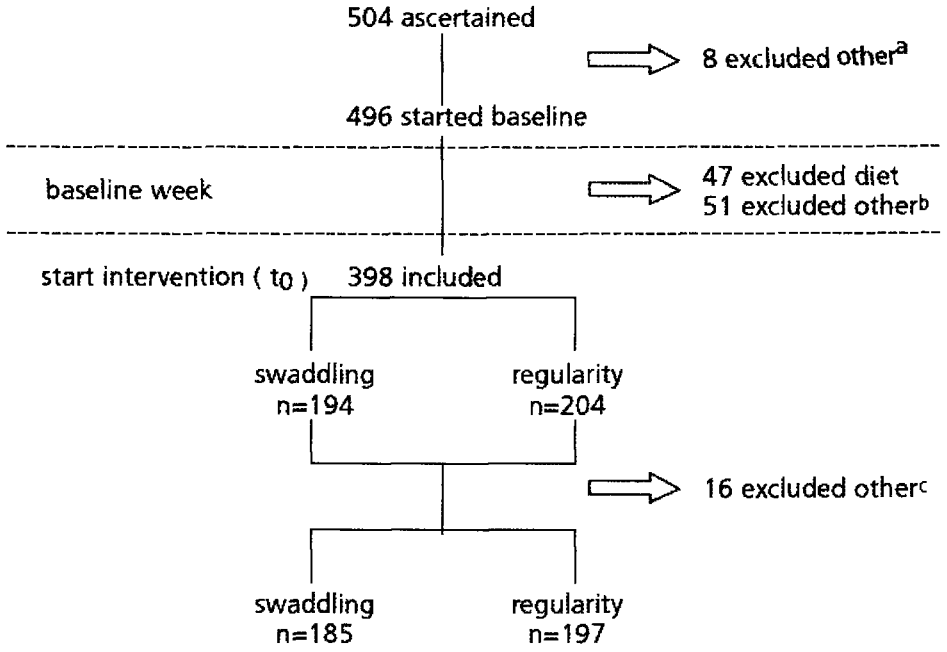


Figure 1. Included and excluded cases.

a. Six not meeting inclusion criteria, 1 admitted to hospital, 1 metabolic disorder.
b. Two not meeting inclusion criteria, 15 sudden unexplained reduction of crying, 6 admitted to hospital, 2 started swaddling, 7 could not identify one-self with the given advises, 4 visited alternative healthcare, 2 treatment of thrush, 3 treatment of reflux, 1 treatment with lactulose, 7 reduction of crying because of malnutrition, 2 transport problem.
c. One not meeting inclusion criteria , 1 admitted to hospital, 1 started swaddling, 12 could not identify one-self with the given advises, 1 visited alternative healthcare.

Demographic Characteristics

Demographic factors were determined from questionnaires (Table 1). First-born infants were overrepresented (56% vs 45.8% in the Dutch population).[20] There were no differences in demographic factors except for paternal age ($P < .006$). Compared with the Dutch population, mothers lived together with partner and infant(s) more often (96% vs 82.8%) and smoked less at time of inclusion (16.6% vs 29.3%).[7,20]

Table 1. Demographic factors.

Parental features	Regularity (n=197)	Swaddling (n=185)	Exclusion diet (n=47)	Exclusion other (n=75)
Age of mother at first live birth (years)[b] -*	29.9 ± 4.42	29.7 ± 4.55	26.5 ± 3.87	28.2 ± 5.01
Age of mother (years)[b] -*	31.9 ± 4.29	32.2 ± 4.63	29.8 ± 4.95	30.3 ± 5.23
Age partner (years)[b] -# *	34.1 ± 5.11	35.6 ± 5.45	31.2 ± 8.00	32.7 ± 5.20
Maternal education[a] -*				
- no education/primary school	3 (1.5)	2 (1.1)	2 (4.3)	11 (15.5)
- lbo/mavo[1]	40 (20.3)	44 (23.8)	10 (21.3)	17 (23.9)
- havo/vwo/mbo[2]	69 (35.0)	73 (39.5)	26 (56.5)	25 (35.2)
- hbo/university[3]	85 (43.1)	66 (35.7)	8 (17.4)	18 (25.4)
Paternal education[a]				
- no education/primary school	4 (2.1)	1 (0.6)	1 (2.2)	3 (4.5)
- lbo/mavo[1]	44 (22.9)	45 (25.0)	17 (37.8)	14 (21.2)
- havo/vwo/mbo[2]	82 (42.7)	61 (33.9)	18 (40.0)	23 (34.8)
- hbo/university[3]	62 (32.3)	73 (40.6)	9 (20.0)	26 (39.4)
Marital status[a] -*				
- living together with partner and infant(s)	188 (95.9)	175 (95.1)	44 (95.7)	65 (95.6)
Maternal smoking indoors at time of interview[a]*				
- yes	29 (14.7)	30 (16.2)	13 (28.3)	11 (16.9)
Maternal smoking indoors at time of interview[a]				
- 1-10 cigarettes/day	6 (50.0)	9 (81.8)	4 (50.0)	3 (50.0)
Maternal smoking during pregnancy[a]				
- yes	32 (16.2)	29 (15.8)	11 (23.4)	12 (18.5)
Maternal smoking during pregnancy[a]-*				
- 1-10 cigarettes/day	23 (76.7)	24 (82.8)	5 (55.6)	8 (72.7)
Paternal smoking indoors at time of interview[a] -*				
- yes	17 (8.9)	14 (7.8)	10 (21.3)	4 (6.3)
Child features				
Gender[a] - boy	100 (50.8)	101 (54.6)	25 (53.2)	44 (62.0)
Birth order[a] - first born	117 (59.4)	91 (49.2)	26 (55.3)	46 (64.8)
Parity[a] - singleton	184 (93.4)	178 (96.2)	45 (95.7)	66 (93.0)
Age of infant at enrollment (weeks)[b]	7.9 ± 2.52	8.0 ± 2.53	8.5 ± 2.43	7.9 ± 2.97
Birth weight (gram)[b]	3338.0 ± 602.34	3430.6 ± 552.17	3422.4 ± 612.29	3405.2 ± 543.92
Birth length (cm)[b]	50.2 ± 2.63	50.6 ± 2.71	50.4 ± 3.68	50.7 ± 2.55
Pregnancy duration (weeks)[b]	39.4 ± 2.03	39.6 ± 1.63	39.6 ± 1.84	39.4 ± 1.81
Delivery duration (hours)[b] -*	11.9 ± 12.64)	12.1 ± 12.60	7.8 ± 7.73	13.7 ± 15.08
Contraction of labour (min)[b]	42.5 ± 51.93)	39.8 ± 40.59	31.0 ± 31.02	53.0 ± 75.96
Crying/24hrs as indicated by parents (hours)[b]	5.5 ± 2.89)	6.0 ± 3.72	6.22 ± 2.70	6.71 ± 4.23

1. Lower technical and vocational training and lower general secondary education
2. Intermediate vocational training and advanced secondary education
3. Higher vocational education (college education) and university

#Significant difference between regularity and swaddling
* Significant difference between inclusion and exclusion diet

[a] Counts (%)
[b] Mean ± SD

Cry Measurements

The mean duration of crying and fussing as calculated from the diaries in the baseline week was 4.37 hours/24 hours (SD 1.79), crying was 2.47 hours/24 hours (SD 1.38), and fussing was 1.90 hours/24 hours (SD 1.14). At t0 parents reported a mean duration of crying of 5.76 hours/24 hours (SD 3.31). Of all infants whose parents reported that their infant cried more than 3 hours a day, 32.3% actually did so according to the diary. At t0 there was no significant difference in crying, fussing, crying/fussing, and sleeping between the two intervention groups.

The reduction in crying in the total group after the first intervention week was 62.1 minutes/24 hours (41.9%) and after 2 weeks 70.7 minutes/24 hours (49.5%). At week 8, stabilizing up to the end of the intervention period, the infants cried 36.4 minutes/24 hours, which is a 75% reduction compared with the amount of crying during the baseline.

During the first week of intervention the amount of crying differed between both study groups. In the R-group, the amount of crying increased by 20 to 25 minutes on the first day, and then, on the following days decreased by about 11 minutes. In contrast, in the RS-group on the first day the amount of crying decreased by 30 to 40 minutes (Figure 2).

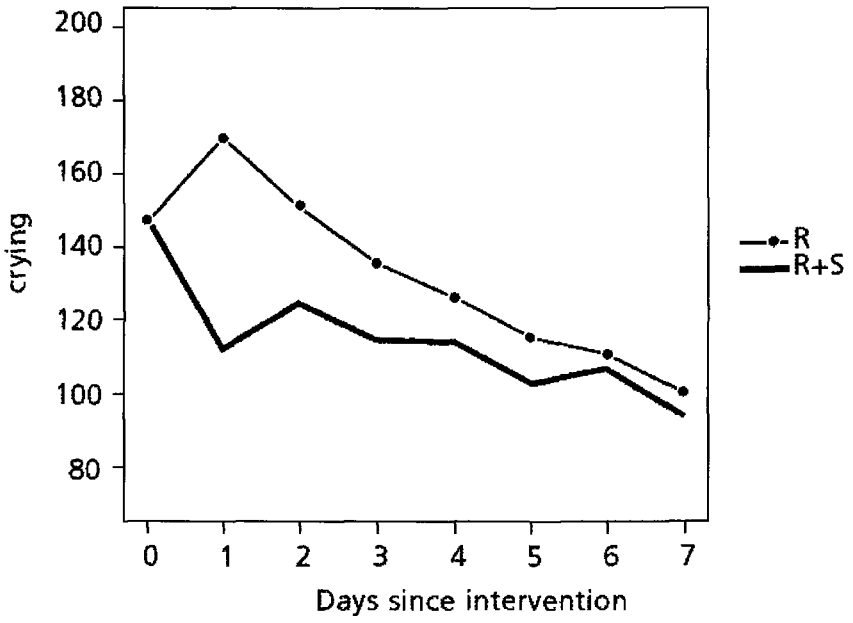


Figure 2. Average amount of crying (minutes/24 hours) during the first intervention week.

After 7 days there was no significant difference (Mann- Whitney *U* test).
In the whole study group, there was no significant difference in decrease of crying between the R- and the RS-group in the total intervention period (Table 2). However, the success of the two interventions differed with age: Infants 1 to 7 weeks of age at randomisation (n = 145, 38%) benefited significantly more from being swaddled, whereas infants 8 to 13 weeks of age at randomisation (n = 237, 62%) benefited significantly more from regularity instructions without swaddling (OR 0.065; 95% CI 0.023;0.107) (Figure 3). The group differences within each age category were approximately 10 minutes. Subgroup analyses between excessive (>3 hours/24 hours for at least 3 days a week) and non-excessive crying did not show a significant difference between the R- and RS-group in the total intervention period. Furthermore, in all (sub) groups R and RS did not differ in amount of fussing (data not shown).

Table 2. Parameter estimates linear mixed model for crying (minutes/24 hours), in which response is adjusted by the covariate baseline (logarithmic transformation).

	Corrected for age baby (wks)		Corrected for other variables*	
	Value	95% CI	Value	95% CI
Intercept	2.44	2.043;2.85##	3.67	2.76;4.58##
Week	-0.0105	-0.12;-0.09##	-0.087	-0.121;-0.052##
Group - RS	-0.0104	-0.242;0.0338	-0.091	-0.232;0.049
Crying	0.4251	0.344;0.5062##	0.378	0.291;0.465##
Week:Group-RS	0.0051	-0.016;0.026	0.006	-0.015;0.027

* Corrected for age of the baby (wk), intervention area, birth weight (g), sex, age mother at first live birth (y), age mother at time of interview (y), age father at time of interview (y), type of feeding, health perception of mother, birth by suction, smoking during pregnancy, current smoking behaviour, General Health Questionnaire total score.
^ $p < .05$
\# $p < .001$

Cry Perception Scale

Comparing the baseline week and week 12, in the parent's perception the crying was less worrisome ($P < .000$). No differences were found between the R- and the RS-group. Subgroup analyses between excessive and non-excessive crying and between younger and older infants showed similar results.

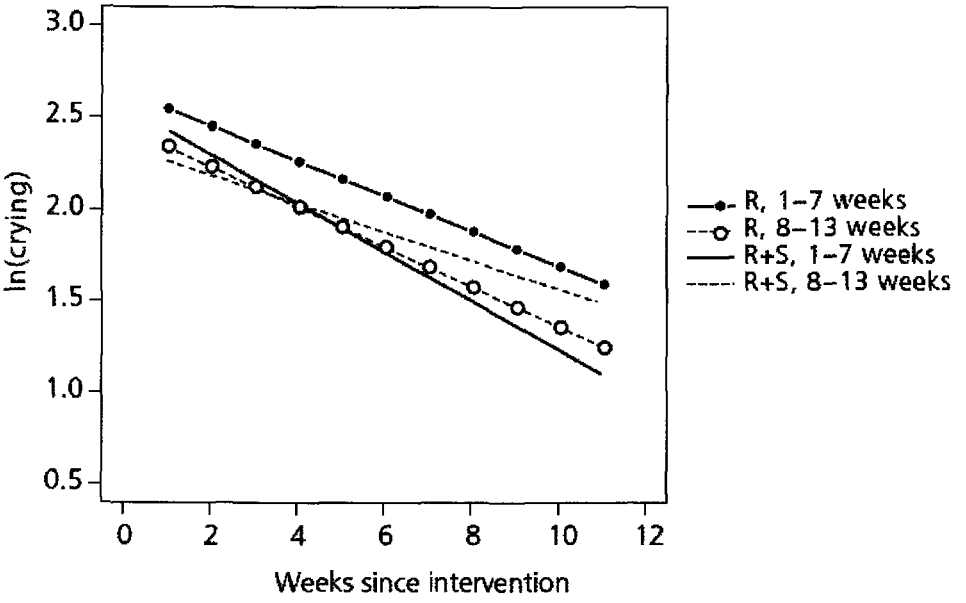


Figure 3. Decrease in amount of crying (minutes/24 hours) during the total intervention period for both age groups (logarithmic transformation).

Discussion

Excessive infant crying occurs often and can lead to serious sequellae.[3-6] Therefore a randomised trial was designed in which two strategies were compared that could both be easily incorporated in the working practices of well-baby clinics, thus offering possibilities for secondary prevention. As it is the parental perception of the crying that leads to frustration and undesirable reactions, parental estimate of the amount of crying was used as the most important inclusion criterion. An unambiguous intervention for excessive crying has not yet been described, although some (non-) effective strategies have.[21-23] Barr et al demonstrated in a randomised controlled trial that one aspect of behaviour modification, carrying the baby, is ineffective.[7] Three systematic reviews show insufficient evidence for simethicone, dicyclomine, methyl scopolamine, lactase enzymes, soy-based formula, casein hydrolysate milk, low lactose milk, sucrose solution, herbal tea, and behavioural modification.[21-23] The use of whey hydrolysate milk does seem effective for subgroups and in primary care settings.[17] In 2% to 5%, excessive crying can be explained by cow's milk protein intolerance.[24,25] Behaviour modification studies differ widely in methods and results and are of course not double-blinded. One study indicates that regularity and stimulus reduction is an effective strategy.[9] Wolke et al compared empathic telephone calls with a behavioural modification similar to ours and a third control group.[9] Although the study was quite small (n = 92) and the included infants were

older (1-5 months of age), the empathy group showed a decrease in crying and fussing of 37.5% after 3 months compared with the behavioural group, 51.2%, and the control group, 35.2%. These data and several advice items concerning prevention of excessive crying by Hofacker et al[10] and Blom[8] were used to construct our intervention of regularity and stimulus reduction.

Possible Limitations of the Study

In the study we chose not to include a "care as usual" intervention group as there is no standardized care in the Netherlands for excessively crying infants. Participating in a study with 24-hour diaries and telephone calls is already incompatible with "care as usual" as the effect of extra attention and empathy cannot be controlled for adequately. Therefore our research question was whether or not swaddling has added benefit to offering a form of behaviour modification of regularity in baby care and stimulus reduction. Another unavoidable drawback is that double-blinding was not possible in a behavioural intervention study.

Strengths

Both interventions were well accepted by the parents, and healthcare workers were positive about the ease of implementation in a well-baby setting. Eleven of 16 parents who switched from the R- to the RS-group after week 5 had a preference for swaddling before randomisation and one parent had no preference. One of the 2 parents who switched from RS- to the R-group expressed doubts about swaddling before randomisation. The sharp increase in crying on the first day after starting the intervention in the R-group can be interpreted as a token of protest of the infants. After only a few days the parent-infant couples seem to adjust to the new routine. Parents should be informed about this initial increase in crying by the infant, to prevent needless distress.

Swaddling

Swaddling is an ancient technique by which movements of children are limited, by tightly winding cloths around them.[26] Research in experimental settings demonstrates that the arousal function is influenced by swaddling, which promotes sleep.[27,28] There is evidence, that swaddling influences arousal and could be a preventive measure for cot death.[27,29] However, swaddling is not uncontroversial, especially in relation to developmental dysplasia of the hip.[30] Participating infants were screened for hip dysplasia, and the method of swaddling used allowed normal leg flexion and abduction. The method also allowed normal chest excursion necessary for unimpeded respiration. Instructions were given to temporarily stop swaddling in case of fever.[31,32] A swaddled infant who is able to turn to prone has an increased risk of cot death.[33] Swaddling should therefore be only applied in infants unable to turn when swaddled and no older than 6 months of age. The young infants (1-7

weeks of age at randomisation) showed a significantly greater decrease in crying during the 11 weeks of intervention if they were swaddled (approximately 12 minutes/24 hours). The older infants (8-13 weeks of age at randomisation) showed a significantly greater decrease in crying in the regularity group without swaddling (approximately 9 minutes/24 hours). Swaddling decreases the infants' level of reactivity through motor restraint. Perhaps this is a more important factor in the younger group. It would therefore be worthwhile to extend this research to the group of premature and small-for-gestational-age infants who are known for their jitteriness.

The natural course of excessive crying is favourable. However, considering the anxiety it causes and the serious sequelae especially in relation to child abuse, it is appropriate to offer parental support. Both interventions offered were well accepted by parents and healthcare workers, and easily implemented in a well-baby clinic setting. Overall, no significant difference was found in the decrease of crying in both intervention groups. Although at the first day of intervention a regular pattern of baby care increases crying, whereas crying was reduced when the infants were additionally swaddled, this difference disappeared after one week. Swaddling may be tried as a supplementary intervention for excessive crying in infants <8 weeks of age, but the effect is modest.

Trial Registration
ISRCTN assigned: ISRCTN18400679
Date Assigned: 20-Dec-05

References

1. Crying in babies. In: Brugman E, Reijneveld SA, Hollander-Gijsman ME den, Burgmeijer RJF, Radder JJ. Gauging in the Child Health Care 1997-1998. Leiden: TNO Prevention and Health; 1999:41-5
2. Wessel, MA, Cobb JC, Jackson EB, Harris GS, Detwiler AC. Paroxysmal fussing in infancy, sometimes called "colic." Pediatrics 1954;94:322-32
3. Karp H. The "fourth trimester": a framework and strategy for understanding and resolving colic. Contemporary Pediatrics® February 2004
4. Reijneveld SA, Wal MF van der, Brugman E, Hira Sing R, Verloove- Vanhorick SP. Infant crying and abuse. Lancet 2004;364:1340-2
5. Weston J. The pathology of child abuse. In: Helfer R, Kempe, C, eds. The Battered Child. Chicago: University of Chicago Press; 1968
6. Holliday-Hanson ML, Barr RG, Trent RB. Does the age-related pattern of shaken baby syndrome (SBS) correspond to the age-related pattern of early crying? Pediatr Res 2001;49(suppl):19A

7. Barr RG, McMullan SJ, Spiess H, Leduc DG, Yaremko J, Barfield R, et al. Carrying as colic "therapy": a randomized controlled trial. Pediatrics 1991;87:623-30
8. Blom MA. Crying and Restlessness in Babies. A Parent's Guide to Natural Sleeping. Edinburgh: Floris Books; 2005
9. Wolke D, Gray P, Meyer R. Excessive infant crying: a controlled study of mothers helping mothers. Pediatrics 1994;94:322-32
10. Hofacker N von, Papousek M, Jacubeit T, Malinowski M. Rätsel der Säuglingskoliken. Ergebnisse, Erfahrungen und therapeutische Interventionen aus der "Müncher Sprechstunde für Schreibabies." Monatsschr Kinderheilkd 1999;147:244-53
11. Barr RG, Kramer MS, Boisjoy C, McVey-White L, Pless IB. Parental diary of infant cry and fuss behaviour. Arch Dis Child 1988;63:380-7
12. Lester BM, Boukydis CFZ, Garcia-Coll CT, Holl WT, Peucker M. Infantile colic: cry characteristics, maternal perception of cry, and temperament. Infant Behav Dev 1992;15:15-26
13. Huizink AC, Mulder EJH, Robles de Medina PG, Visser GHA, Buitelaar JK. Is pregnancy anxiety a distinctive syndrome? Early Hum Dev 2004;79:81-91
14. Ark LA van der, Marburger D, Mellenbergh GJ, Vorst HCM, Wald F. Revised Profile of Mood States (POMS – r). Manual and Justification [In Dutch]. Nijmegen: Berkhout Nijmegen BV; 1995.
15. Koeter MWJ, Ormel J. General Health Questionnaire [Dutch Translation]. Lisse: Swets & Zeitlinger BV; 1991
16. Sonderen E van. Social Support List – Interactions (SSL – I) and Social Support List – Discrepancy (SSL – D). Manual [In Dutch]. Groningen: University of Groningen; 1993
17. Lucassen LB, Assendelft WJ, Gubbels LW, Eijk JT van, Douwes AC. Infantile colic: crying time reduction with a whey hydrolysate; a double blind, randomized placebo-controlled trial. Pediatrics 2000;106:1349-54
18. Rautava P, Helenius H, Lehtonen L. Psychosocial predisposing factors for infantile colic. BMJ 1993;307:600-4
19. Høgdall CK, Vestermark V, Birch M, Plenov G, Toftager-Larsen K. The significance of pregnancy, delivery and postpartum factors for the development of infantile colic. J Perin Med 1991;19:251-7
20. Central Bureau for Statistics (CBS). Birth: numbers of gender, birth order (first, second child etc. from mother) and legitimacy. Also number of infants born dead and multiparity. 1950-2002 [In Dutch]. Voorburg; 2003
21. Garrison MM, Christakis DA. A systematic review of treatments for infantile colic. Pediatrics 2000;106:184-90
22. Lucassen PLBJ, Assendelft WJJ, Gubbels JW, Eijk JTM van, Geldrop WJ van, Knuistingh Neven A. Effectiveness of treatments for infantile colic; systematic review. BMJ 1998;316:1563-69
23. Wade S, Kilgour T. Infantile colic. BMJ 2001;323:437-40

24. Crowncroft NS, Strachan DP. The social origins of infantile colic: questionnaire study covering 76.747 infants. BMJ 1997;314:1325-8
25. Taubman B. Parental counseling compared with the elimination of cow's milk or soy protein for the treatment of infant colic syndrome: a randomized controlled trial. Pediatrics 1988;81:756-61
26. Lipton EL, Steinschneider A, Richmond JB. Swaddling, a child care practice: historical, cultural, and experimental observations. Pediatrics 1965;35:521-67
27. Gerard CM, Harris KA, Thach BT. Spontaneous arousals in supine infants while swaddled and unswaddled during Rapid Eye Movement and Quiet Sleep. Pediatrics 2002;110:e70
28. Franco P, Seret N, Hees JN van, Scaillet S, Groswasser J, Kahn A. Influence of swaddling on sleep and arousal characteristics of healthy infants. Pediatrics 2005;115:1307-11
29. Gerard CM, Harris KA, Thach BT. Physiologic studies on swaddling: an ancient child care practice, which may promote the supine position for infant sleep. J Pediatr 2002;141:398-404
30. Kutlu A, Memik R, Mutlu M, Kutlu R, Arslan A. Congenital dislocation of the hip and its relation to swaddling used in Turkey. J Ped Ortho 1992;12:598-602
31. Yurdakok K, Yavuz T, Taylor CE. Swaddling and acute respiratory infections. Am J Public Health 1990;80:873-5
32. Gestel JPJ van, L'Hoir MP, Berge M ten, Jansen NJG, Plötz FB. Risks of ancient practices in modern times. Pediatrics 2002;110:e78
33. Fleming PJ, Gilbert R, Azaz Y, Berry PJ, Rudd PT, Stewart A, et al. Interaction between bedding and sleeping position in the sudden infant death syndrome: a population based case-control study. BMJ 1990;301:85-9

Characteristics of excessively crying infants who react positively to a hypoallergenic diet

5

Bregje E. van Sleuwen
Louise Berkhout
Monique P. L'Hoir
Tom W.J. Schulpen
Wietse Kuis
Yolanda Meijer
Adèle C. Engelberts

Submitted for publication

Abstract

Introduction
Of 496 infants, who participated in a randomised trial on the effect of behaviour modification with and without swaddling on excessive crying (2001-2003), 47 (10%) showed a strong reduction of crying after one week hypoallergenic diet only (baseline responders, BR). In a follow-up study, we identified how many of these children received a re-challenge, re-elimination test and were diagnosed cow's milk allergic by an open or double blind provocation test. Furthermore, we studied which determinants discriminate between favourable and non-favourable reactions to hypoallergenic diet.

Methods
All 47 BR who were excluded in the swaddling study were contacted for telephone questionnaire in 2005, around 2.5 years after the baseline study, and their well-baby clinic records were collected. These BR were compared to 382 children who were included in the swaddling study as they did not react positively to a hypoallergenic diet (baseline non-responders, BNR).

Results
Of 47 BR, we located 35 (74%) infants with a mean age of 35 months. When comparing the baseline data, BR parents reported more maternal asthma, paternal hay fever, and parental atopy, they smoked more often than BNR parents, they less often exclusively breastfed, and they were less stressed during first trimester of pregnancy. Postnatal smoking was only in the BR group associated with excessive crying. Only 12 (34.3%) of the infants followed the advised re-challenge, re-elimination test at their well-baby clinic of whom 6 had proven cow's milk allergy. At the age of 35 months, only three infants received no cow's milk of whom one followed a hypoallergenic diet.

Conclusion
We conclude that 12 of 35 excessively crying infants who reacted favourable after one-week hypoallergenic diet, followed the advice to return to the well-baby clinic for a re-challenge, re-elimination test and, of these, only 6 were diagnosed cow's milk allergic. At follow-up, 32 of the 35 infants had cow's milk in their diet. At well-baby clinics, more effort should be put into diagnosing cow's milk allergy with a double blind provocation test.

Keywords

hypoallergenic diet, excessive crying, colic, cow's milk allergy, randomised trial

Introduction

About 2%-2.5% of newborn infants show hypersensitive reactions to cow's milk in the first year of life.[7,22] In the Netherlands a prospective cohort study revealed a 2.5% incidence of cow's milk allergy, diagnosed by means of two positive cow's milk elimination/challenge tests in 1158 unselected infants; in colicky infants (more than 2h daily crying for more than 2 weeks) the incidence of diagnosed cow's milk allergy was 6%.[24] A recent study on the incidence of cow's milk allergy in excessively crying infants admitted to a hospital reveals a 2% incidence of diagnosed cow's milk allergy.[17]

Various causes are described in the literature for excessive crying, such as cow's milk allergy, gastro-oesophageal reflux, infections, or various rare conditions, but in most cases excessive crying remains unexplained.[15,30] Literature into the role of food allergy in the pathophysiology of colic is controversial.[6]

Double blind placebo controlled food challenges (DBPCFC) are considered to be the gold standard in the diagnosis of food related symptoms.[16,21] However, quite often this level of evidence is lacking and parents and professionals assume that cow's milk allergy was the reason behind crying.

In the USA, one fourth of all households have altered their diets as at least one member is perceived to have food allergies.[26] Overdiagnosis of food allergy and intolerance has led to malnutrition, eating disorders, psychosocial problems, and family disruption.[23]

In this study, we compared infants who, based on baby day diaries and/or parental perception of crying behaviour, responded positively to a 1-week cow's milk elimination diet, to a group of infants whose crying did not reduce after the elimination diet.

Methods

The goal of this study is to define differences between infants who, according to their parents, reacted positively to a hypoallergenic diet and those who did not in a group of 398 excessively crying infants. All infants were offered hypoallergenic diet for a period of a week as a baseline measure before inclusion in a randomised trial to compare the effect on excessive crying of behaviour modification with and without swaddling.[25] If crying reduced dramatically after the diet, the infant was not included in the randomised trial. However, this does not mean a cow's milk allergy has been proven. By means of a limited follow-up, we determined:

1. how many children received a re-challenge, re-elimination test;
2. how many infants were diagnosed as allergic;

and which determinants discriminate between;

3. infants with favourable and non-favourable reactions to hypoallergenic diet, and;
4. infants that were indeed allergic and those in which allergy was not diagnosed.

Design

Four hundred ninety six cases were ascertained for the randomised trial on the effect of behaviour modification with and without swaddling on excessive crying (Figure 1).[25] A standardized case definition with the modified Wessel's criteria of excessive crying (i.e., crying > 180 min/24hr for at least 3 days a week) was used for recruitment.[2,29] The perception of the parents and/or doctors about the amount of crying was decisive for participation. The actual frequency and duration of crying was established in a baseline week by a 24-hour diary.[3]
Infants were referred from February 2001 – March 2003 by well-baby clinics, general practitioners, (children's) hospitals, or by self-referral. Inclusion criteria were healthy infants with a maximum age of 12 weeks and 6 days and a minimal gestational age of 32 weeks.
In the baseline week all infants received a hypoallergenic diet (Frisolac Allergycare, an extensively hydrolysed casein-based formula, or, in case of breastfeeding, a maternal diet free of milk, egg, (shell) fish, nut products and seeds). All infants in which crying had been reduced to less than 120 min/24hr from day three/four of the baseline week onwards and/or in their parents perception were excluded from the randomised trial and enrolled in the presumed allergic follow up study. From November 2004 – April 2005, these infants, then aged about 2.5 years, were contacted for follow-up by means of a telephone questionnaire and their well-baby clinic records were collected.
The Medical Ethical Committee of the University Medical Centre Utrecht approved the study. All parents gave informed consent.

Measurements, obtained during the first visit

In the baseline week parents recorded infant behaviour for 7 consecutive days in a *24-hour* diary.[3,25] Maternal perception of infant crying and the maternal reaction to the cry were recorded twice during the baseline week in the *Cry Perception Scale*.[13,25] During the parental interview carried out before the start of baseline week, questions about allergy, diagnosis and feeding behaviour were asked, as well as family atopy and various medical and psychological measures.[1,8,11,14,18,25,27] Data related to the Pregnancy Related Anxiety Questionnaire – Revised (PRAQ-R) are given.[10] The PRAQ-R is an abbreviated version of the PRAQ, which measures specific fears and worries related to pregnancy. A high score means more maternal anxiety and pregnancy related fears.

Statistical analysis

All data were analysed in Statistical Package for Social Science (SPSS Inc., Chicago Ill.) for Windows 12.0. T-tests, Pearson χ^2 tests, Mann-Whitney U tests and ANOVA's were used to analyse the demographic data and the Cry Perception Scale.

Cry features

From the diary we calculated the total and mean duration of crying and fussing (min/24hr).

Results

Response

In the study 496 infants entered the baseline week, 51 parents (10.3%) decided to stop participation during this week (Figure 1).[25] After a completed baseline week with hypoallergenic diet 47 infants (10.6%) were excluded as they responded positively to the diet. Thirty three infants had reduced their crying to less than 120min/24hr, and 12 infants still cried >120min/24hr according to the baby day diary, but it was less than before and their parents did not perceive their crying as excessive anymore (of 2 infants, the baby day diaries were not complete). These 47 were considered as baseline reducers (BR) possibly related to the hypoallergenic diet. Those that did not reduce crying during the baseline week were named baseline non-reducers (BNR). Of the 47 BR, we were able in 2005 to locate 35 for follow-up (74.4%) of whom we collected 30 well-baby clinic records (63.8%). All contacted parents acquiesced to the interview.

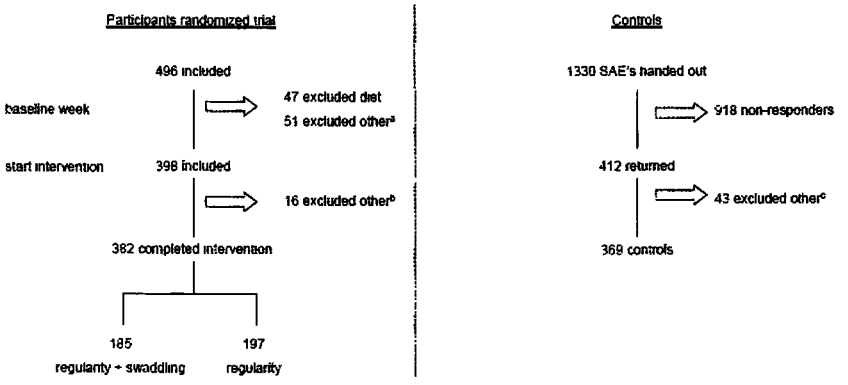


Figure 1. In- and excluded cases.

a. 2 not meeting inclusion criteria, 15 sudden unexplained reduction of crying, 6 admitted to hospital, 2 started swaddling, 7 could not identify one self with the given advice, 4 visited alternative health care, 2 treatment of thrush, 3 treatment of reflux, 1 treatment with lactulose, 7 reduction of crying because of malnutrition, 2 transport problem
b. 1 not meeting inclusion criteria, 1 admitted to hospital, 1 started swaddling, 12 could not identify one self with the given advice, 1 visited alternative health care
c. 23 cried excessively, 10 not meeting inclusion criteria, 10 incomplete questionnaires

Demographic characteristics

Demographic characteristics reveal that BR mothers were younger at first live birth compared to the mothers in the BNR group and fathers were younger as well (Table 1). Furthermore, the BR mothers were less often university-educated.

Cry measurements

The mean duration of crying and fussing for BNR infants as calculated from the diaries in the baseline week was 262.97 min/24hr (SD 120.22) (crying 153.04 min/24hr (SD 92.26) and fussing 111.07 min/24hr (SD 76.05)), for BR infants the mean duration of crying and fussing was 172.54 min/24hr (SD 84.11) (crying 97.17 min/24hr (SD 58.86) and fussing 76.09 min/24hr (SD 55.87)). At start baseline (day 2) the BR and BNR groups did not significantly differ (p = 0.163). During the baseline week the crying of the BR (n=47) shows a greater decline than of the BNR (Figure 2). At day 7 of the baseline week the BNR had a mean duration of crying of 138,37 min/24hr (SD 117,5), and the BR cried 54,59 min/24hr (SD 51,46; p = 0.000).

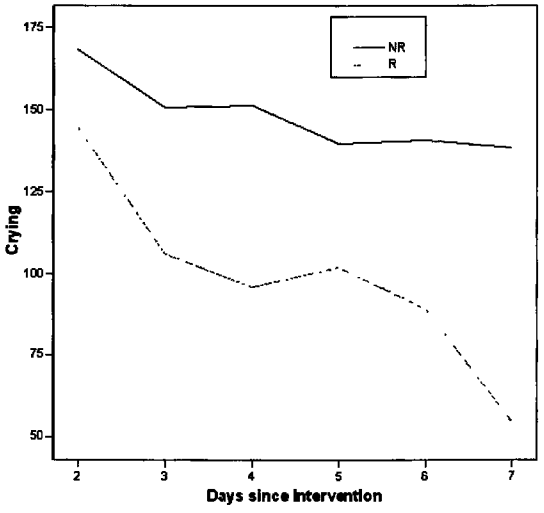


Figure 2. Average amount of crying (min/24 hr) during the first intervention week for the baseline responders (R) and the baseline non-responders (NR).

Features study group

Table 2 shows the family history of atopy and several other characteristics for study infants. The parents of BR reported significantly more maternal asthma (p= 0.007) and paternal hay fever (p= 0.011). Furthermore, significantly more atopy was present in one as well as in both parents compared to the BNR group.

Table 1. Demographic characteristics.

	Baseline Responders (BR) (n=47)	Baseline Non-Responders (BNR) (n=382)	OR (95% CI)
Parental features			
Age of mother at first live birth (years)[b] *	26.53 ± 3.87	29.81 ± 4.48	0.845 (0.786 - 0.909)
Age of mother (years)[b] *	29,78 ± 4,95	32.06 ± 4.46	0.894 (0.835 - 0.958)
*Age partner (years)[b] **	*31,87 ± 5.63*	*34.8 ± 5.33*	*0.888 (0.831 - 0.950)*
Maternal education[a] *			
- no education/primary school/lbo/mavo[1]	12 (26.1)	89 (23.3)	1
- havo/vwo/mbo[2]	26 (56.5)	142 (37.2)	1.358 (0.652 - 2.828)
- hbo/university[3]	8 (17.4)	151 (39.5)	0.393 (0.155 - 0.998)
Marital status[a]			
- living together with partner and infant(s)	44 (95.7)	363 (95.5)	0.971 (0.217 - 4.342)
Child features			
Gender[a] - boy	25 (53.2)	201 (52.6)	0.977 (0.533 - 1.793)
Birth order[a^] - first born	26 (55.3)	208 (54.5)	0.966 (0.525 - 1.776)
Age of infant at enrolment (weeks)[b]	8.47 ± 2.43	7.98 ± 2.2	1.081 (0.957 - 1.220)
Birth weight (gram)[b]	3422.43 ± 612.29	3382.73 ± 579.75	1.000 (1.000 - 1.001)
Pregnancy duration (weeks)[b]	39.62 ± 1.84	39.49 ± 1.85	1.039 (0.878 - 1.231)
Delivery duration (hours)[b] *	7.8 ± 7.73	11.97 ± 12.60	0.955 (0.915 - 0.997)

1. Lower technical and vocational training and lower general secondary education
2. Intermediate vocational training and advanced secondary education
3. Higher vocational education (college education) and university

a. Counts (percentages)
b. Mean ± SD

* Significant difference between baseline non-responders and baseline responders

Table 2. Parental and infant characteristics.

Parental features	Baseline Responders (n=47)	Baseline Non-Responders (n=382)	OR (95% CI)
Allergies reported by mother[a]			
- yes	17 (36.2)	93 (24.3)	1.761 (0.929 – 3.337)
Maternal eczema (as a child)[a]			
- yes	14 (29.8)	81 (21.2)	1.577 (0.805 – 3.086)
Maternal hay fever[a]			
- yes	14 (29.8)	84 (22.0)	1.505 (0.770 – 2.942)
Maternal asthma/bronchitis[a] *			
- yes	17 (36.2)	70 (18.3)	**2.526 (1.320 – 4.833)**
Maternal food allergy[a]			
- yes	3 (6.4)	35 (9.2)	0.676 (0.200 – 2.290)
Paternal eczema (as a child)[a]			
- yes	7 (14.9)	62 (16.2)	0.903 (0.387 – 2.109)
Paternal hay fever[a] *			
- yes	16 (34.0)	67 (17.5)	**2.427 (1.256 – 4.688)**
Paternal asthma/bronchitis[a]			
- yes	11 (23.4)	60 (15.7)	1.640 (0.791 – 3.400)
Paternal food allergy[a]			
- yes	6 (12.8)	21 (5.5)	2.516 (0.960 – 6.590)
Atopy present in one parent[a]*			
- yes	37 (78.7)	243 (63.6)	**2.116 (1.021 – 4.387)**
Atopy present in both parents[a]*			
- yes	17 (36.2)	80 (20.9)	**2.139 (1.123 – 4.073)**
Child features			
Type of feeding[a]			
- Exclusively breastfed	14 (29.8)	160 (41.9)	
- Wholly/partially bottle-fed	33 (70.2)	222 (58.1)	1.699 (0.880 – 3.278)
Throwing up after feeding [a]			
- 1-2 times a day/always	11 (23.4)	105 (27.6)	1.250 (0.613 – 2.546)
Wheezing after feeding [a]			
- 1-2 times a day/always	4 (9.5)	57 (15.1)	1.697 (0.583 – 4.940)
Red spots after feeding [a]			
- 1-2 times a day/always	5 (11.3)	23 (6.1)	0.511 (0.184 – 1.420)
Often stomach ache when crying[a] *	15 (34.1)	191 (52.0)	**0.477 (0.247 – 0.919)**
Often problems with defecation[a]	16 (34.8)	120 (31.4)	1.164 (0.612 – 2.217)
Often unsoothable during crying[a]	27 (58.7)	213 (56.1)	0.898 (0.482 – 1.670)
Often flatus during crying[a]	28 (60.9)	189 (49.9)	1.564 (0.837 – 2.923)
Often red face during crying[a]	38 (82.6)	307 (81.0)	1.114 (0.498 – 2.490)
Often swollen belly when crying[a]	18 (40.0)	139 (39.4)	1.026 (0.545 – 1.934)

a. Counts (percentages)

* Significant difference between baseline non-responders and baseline responders

Psychological features during pregnancy

In first trimester of pregnancy the mothers of BR infants showed significantly less anxiety than the BNR group as shown by several items of the Pregnancy Related Anxiety Questionnaire – Revised (PRAQ-R) (data not shown).[10]

Cry Perception Scale

At start of the randomised trial, BR parents rated their infants as significantly more sick on the healthy/sick item compared to BNR parents (OR 1.254, 95% CI 1.036 – 1.518).

Results follow-up interview

At time of follow-up, BR (n=35) had a mean age of 34.5 months (SD 7.75), 60% of them were boys and 62.9% were first-born (Table 1). Seven (21.2%) mothers and 12 (36.4%) fathers smoked at time of follow-up, 4 mothers and 10 fathers smoked indoors (Table 3).

- Family history

Twelve infants of the BR (34.3%) had no brothers or sisters at time of follow-up. Of all brothers or sisters, 9 (39%) had eczema, 1 (4.3%), hay fever, 3 (13%), asthma, and 7 (30.4%) had a food allergy as reported by their parents.

- Diagnosis of atopy

As reported by the parents, in only 12 (34.3%) infants the Dutch well-baby clinic protocol to diagnose cow's milk allergy (an open cow's milk elimination/re-challenge/re-elimination test) was performed, in eleven this was confirmed by the well-baby clinic records (one missing). In only 6 the cow's milk allergy was diagnosed, which was confirmed in five well-baby clinic records (one could not be traced). No differences were found on parental and infant characteristics and parental smoking between infants that were indeed allergic and those in whom allergy was not diagnosed, except for the factor 'swollen belly when crying'.

At time of follow-up, parents of 5 (14%) infants reported an ongoing cow's milk allergy; in four of them the well-baby clinic protocol (an open cow's milk elimination/re-challenge/re-elimination test) confirmed a 'cow's milk allergy'. Only one of the five still follows a hypoallergenic diet and also has an egg, soy and peanut allergy, as reported by the parents. According to their parents ten of 35 infants have other allergies (eggs, soy, shellfish, (pea-) nuts, wheat, fruit). Of the six infants with a 'cow's milk allergy', four had one or more other allergies.

- Feeding habits

At the time of follow-up, 32 of the 35 infants consume regular cow's milk or (sweetened) cow's milk products, one hypoallergenic formula and 2 soymilk with an average of 335 ml milk products per day. At the average age of 5.8 months and 6.1 months respectively fruit and vegetables were introduced.

Table 3. Parental smoking characteristics.

	Baseline Responders (BR) (n=47 at baseline)	Baseline Non-Responders (BNR) (n=382)	OR (95% CI)
Maternal smoking indoors at time of baseline[a] *			
- yes	13 (28.3)	59 (15.4)	0.464 (0.230 – 0.933)
Maternal smoking during pregnancy[a]			
- yes	11 (23.4)	61 (16.0)	0.624 (0.301 – 1.293)
Paternal smoking indoors at time of baseline[a] *			
- yes	10 (22.7)	31 (8.4)	0.311 (0.140 – 0.689)
Maternal smoking during pregnancy and at time of baseline[a] *			
- yes	10 (21.3)	47 (12.3)	0.507 (0.236 – 1.088)
	Baseline Responders (BR) (n=33 at follow up)		
Maternal smoking at follow up[a]			
- yes	7 (21.2)	-	-
Maternal smoking indoors at follow up[a] *			
- yes	4 (12.1)	-	-
Paternal smoking at follow up[a]			
- yes	12 (36.4)	-	-
Paternal smoking indoors at follow up[a]			
- yes	10 (30.3)	-	-

a. Counts (percentages)

* Significant difference between BNR and BR

Discussion

Food allergy is one of the possible explanations of excessive crying. Literature into the role of food allergy in the pathophysiology of colic is controversial[6] and this study demonstrates that indeed the relationship between food intolerance, allergy and crying is complex. Personal perceptions, either about whether the crying sounds more often as 'sick' or reduction of the crying due to hypoallergenic diet, seem to influence the diagnosis. As both Crowcroft and Taubman have described earlier, only 2-5% of excessive crying is reported by parents or diagnosed by an open elimination, provocation test as cow's milk intolerance.[4,28]

In a group of 445 infants that completed the baseline week in our study, 47 (10.6%) responded to the hypoallergenic diet. It appeared that of 35 infants that were included at follow-up, regretfully only 12 (34%) had been challenged according to well-baby clinic protocol. Only 6 (50%) of those challenged were diagnosed as cow's milk allergic. Actually, even less are presumably atopic, as the gold standard is a DBPCFC. [16,21] The well-baby clinic protocol undoubtedly gives fall-positive results, estimated at 30-68%.[9] On the other hand, a percentage of those not challenged may indeed be atopic.

The fact that family atopy is an effective predictor of allergy in an infant is described in many studies.[5] Especially maternal asthma and paternal hay fever seem to be associated with those children whose colic responds favourably to a hypoallergenic diet. Of interest is that at follow-up almost 40% of their siblings were reported to be suffering from eczema.

Parents might choose an illness or cow's milk allergy as an explanation for excessive crying. They are informed about this relationship by the popular media and such a cause releases them from responsibility: their infant care practices are not related to the crying and a medical intervention such as a hypoallergenic diet changes parental perception about their infant's cry. We suggest that parental attribution could be a possible explanation, but more research is warranted.

During first trimester of pregnancy, interestingly enough the BR mothers are less anxious compared to BNR mothers. Furthermore, both parents are younger, and mothers were less often university educated. This group of parents might be at risk for medicalisation of their infant's excessive crying, because the majority of the BR were not diagnosed as cow's milk allergic but after a week hypoallergenic diet their crying decreased. This is an indication that the attributions of excessive crying given by the mothers might well be different in both groups.

The strength of this follow-up study is that it was prospective. As far as we know, this is the first study that has follow-up information of excessively crying infants that responded to a hypoallergenic diet. Although the numbers in current study are small, 74% of the infants were available for follow-up. When parents in the study group estimated the amount of crying there was not much difference between the estimated amount of crying/fussing and the crying/fussing according to the diary: parents estimated 346 min/24hr while the average duration according to the diary

was 263 min/24hr. Excessively crying infants were excluded from further participation if the actual duration of crying and/or their perception about the crying was less than 120 min/24hr after hypoallergenic diet. We suggest that the seriousness of crying should always be measured in two ways; cry perception by parents and crying measured with a 24-hour diary.[3,13] Only then, can we hope to gain insight into the complex relationship between objective reduction and perceived reduction of crying.

Beside the fact that an open cow's milk elimination challenge test is not the most reliable instrument to diagnose a cow's milk allergy in (excessively crying) infants, only one third of these infants received such a test. Parents postpone or even skip the allergy challenge probably because they are afraid of a relapse of excessive crying. Apparently this fear subsides, as our data show that almost 90% of parents in the BR group give their infants regular cow's milk products at time of follow up. At least more guidance should be given to carry out a food challenge and instead a more reliable test should be considered; the DBPCFC.[16,21]

Earlier Dutch studies have shown that there is a significant association between smoking and excessive crying.[19,20] Excessively crying infants who responded to a week hypoallergenic diet (BR) in our study have significantly more parents who smoked indoors at time of baseline interview compared to BNR and control infants. It is noteworthy that, although in 60% of the BNR group both parents abstained from smoking indoors, in only 24% of BR both parents did so.
A remarkable finding is that the association between smoking and crying thus seems to be limited to the group of infants that responds to hypoallergenic diet and is at higher risk of being atopic due to family history. In the 2000 study by Reijneveld and colleagues, breastfeeding lessened the effect of smoking on infant crying.[20] Due to the smaller numbers in our study, we cannot correct for this. It has been shown that smoking both pre- and postnatally elevates the risk to develop cow's milk allergy with a relative risk of about 2.[12] Conceivably this could explain in part the association found in our study, but further studies are obviously needed.

Conclusion

In our follow-up study of excessively crying infants that responded to hypoallergenic diet we estimated that only a small percentage of all excessively crying infants do so due to cow's milk allergy. In contrast to this, about 11% of infants appear to respond to the diet according to their parents. Although parents were explicitly advised to carry out a food challenge at their well baby clinic, only 34% of the parents did so. More guidance should be given to parents in order to increase the number of infants appropriately diagnosed with cow's milk allergy. As can be expected, if there is a family history of atopy, there is more chance of a favourable response to a hypoallergenic diet. There appear to be subgroups of mothers of excessively crying

infants. Mothers of BR infants more often perceive their infants' crying as sick on the cry perception scale.
Postnatal parental smoking was associated with excessive crying in our study, as in previous studies, but this was limited to those infants that responded to hypoallergenic diet. These last two factors warrant further investigation.

References

1. Ark LA van der, Marburger, D, Mellenbergh GJ, Vorst HCM, Wald F. Revised Profile Of Mood States (POMS – r). Manual and Justification [In Dutch]. Nijmegen: Berkhout Nijmegen BV, 1995
2. Barr RG. Infant crying and colic: it's a family affair: invited commentary. Inf Ment Health J 1995;16:218-20
3. Barr RG, Kramer MS, Boisjoy C, McVey-White L, Pless IB. Parental diary of infant cry and fuss behaviour. Arch Dis Child 1988;63:380-7
4. Crowcroft NS, Strachan DP. The social origins of infantile colic: questionnaire study covering 76 747 infants. BMJ 1997;314:1325-8.
5. Hays T, Wood RA. A systematic review of the role of hydrolyzed infant formulas in allergy prevention. Arch Pediatr Adolesc Med 2005;159:810-6
6. Heine RG. Gastroesophageal reflux disease, colic and constipation in infants with food allergy. Curr Opin Allergy Clin Immunol 2006;6:220-5
7. Heine RG, Elsayed S, Hosking CS, Hill DJ. Cow's milk allergy in infancy. Curr Opin Allergy Clin Immunol 2002;2:217-25
8. Høgdall CK, Vestermark V, Birch M, Plenov G, Toftager-Larsen K. The significance of pregnancy, delivery and postpartum factors for the development of infantile colic. J Perin Med 1991;19:251-7
9. Hospers IC, De Vries-Vrolijk K, Brand P. Double-blind, placebo-controlled cow's milk challenge in children with alleged cow's milk allergies, performed in a general hospital: diagnosis rejected in two-thirds of the children [In Dutch]. Ned Tijdschr Geneeskd 2006;150:1292-7
10. Huizink AC, Mulder EJH, Robles de Medina PG, Visser GHA, Buitelaar JK. Is pregnancy anxiety a distinctive syndrome? Early Hum Dev 2004;79:81-91
11. Koeter MWJ, Ormel J. General Health Questionnaire [Dutch Translation]. Swets & Zeitlinger BV, Lisse, the Netherlands, 1991
12. Kulig M, Luck W, Lau S, Niggemann B, Bergmann R, Klettke U, Guggenmoos-Holzmann I, Wahn U, Multicenter Allergy Study Group, Germany. Effect of pre- and postnatal tobacco smoke exposure on specific sensitization to food and inhalant allergens during the first 3 years of life. Allergy 1999;54:220-8
13. Lester BM, Boukydis CFZ, Garcia-Coll CT, Holl WT, Peucker M. Infantile colic: cry characteristics, maternal perception of cry, and temperament. Infant Behav Dev 1992;15:15-26

14. Lucassen LB, Assendelft WJ, Gubbels LW, Eijk JT van, Douwes AC. Infantile colic: crying time reduction with a whey hydrolysate; a double blind, randomized placebo-controlled trial. Pediatrics 2000;106:1349-54
15. Lucassen PLBJ, Assendelft WJJ, Gubbels JW, Eijk JTM van, Geldrop WJ van, Knuistingh Neven A. Effectiveness of treatments for infantile colic; systematic review. BMJ 1998;316:1563-1569
16. Niggeman B, Beyer K. Pitfalls in double-blind, placebo-controlled oral food challenges. Allergy 2007;62:729-732
17. Nooitgedagt JE, Zwart P, Brand PLP. Causes, treatment and clinical outcome in infants admitted because of excessive crying to the paediatric department of the Isala clinics, Zwolle, the Netherlands, 1997/'03 [In Dutch]. Ned Tijdschr Geneeskd 2005;149:472-7
18. Rautava P, Helenius H, Lehtonen L. Psychosocial predisposing factors for infantile colic. BMJ 1993;307:600-4
19. Reijneveld SA, Lanting CI, Crone MR, Van Wouwe JP. Exposure to tobacco smoke and infant crying. Acta Paediatr 2005;94:217-21
20. Reijneveld SA, Brugman E, Hirasing RA. Infantile colic: maternal smoking as a potential risk factor. Arch Dis Child 2000;83:302-3
21. Sampson HA. Food allergy – accurately identifying clinical reactivity. Allergy 2005;60 (suppl. 79):19-24
22. Sampson HA. Food allergy. Part 1: Immunopathogenesis and clinical disorders. J Allergy Clin Immunol 1999;103:717-28
23. Sampson HA. Food allergy. Part 2: Diagnosis and management. J Allergy Clin Immunol 1999;103:981-9
24. Schrander JJP, Bogart JPH van den, Forget PP, Schrander-Stumpel CTRM, Kuijten RH, Kester ADM. Cow's milk protein intolerance in infants under 1 year of age: a prospective epidemiological study. Eur J Pediatr 1993;152:640-4
25. Sleuwen BE van, L'Hoir MP, Engelberts AC, Busscher WB, Westers P, Blom MA, Schulpen TWJ, Kuis W. Comparison of behavior modification with and without swaddling as interventions for excessive crying. J Pediatr 2006;149:512-7
26. Sloan AE, Powers ME. A perspective on popular perceptions of adverse reactions to food. J Allergy Clin Immunol 1986;78:127-33
27. Sonderen E van. Social Support List – Interactions (SSL – I) and Social Support List – Discrepancy (SSL – D). Manual [In Dutch]. University of Groningen, the Netherlands, 1993
28. Taubman B. Parental counselling compared with the elimination of cow's milk or soy protein for the treatment of infant colic syndrome: a randomized controlled trial. Pediatrics 1988;81:756-61
29. Wessel MA, Cobb JC, Jackson EB, Harris GS, Detwiler AC. Paroxysmal fussing in infancy, sometimes called "colic". Pediatrics 1954;94:322-32
30. Zwart P, Vellema-Goud MG, Brand PL. Characteristics of infants admitted to the hospital for persistent colic, and comparison with healthy infants. Acta Peadiatr 2007;96:401-5

Psychological factors related to the development of infantile excessive crying

6

Bregje E. van Sleuwen
Adèle C. Engelberts
Tom W.J. Schulpen
Wietse Kuis
Monique P. L'Hoir

Submitted for publication

Abstract

Objective

In a randomised trial into the added effect of swaddling on excessive crying, we investigated psychological state and social measures between mothers of infants who cried excessively, and then compared these to control mothers of non-excessively crying infants. Furthermore, we studied the relation between high and low maternal scores (clinical versus non-clinical range) on several psychological measurements and the outcome of the intervention.

Patients and methods

In the randomised trial, 496 infants received a one-week hypoallergenic diet. 47 children reduced their crying to 2 hours or less (DIET+ group) and 51 were excluded for different reasons. After this baseline week, a remaining 398 (1 to 12 weeks old) infants received the intervention (I-group). Furthermore, 369 healthy non-crying infants served as controls.

Results

Mothers of the I-group perceived more negative feelings and less support during pregnancy than mothers of the DIET+ group. Pertaining to current psychological state, the mothers of infants that cried differed significantly from control mothers, but remarkably little within the group itself. The differences found were related to social support items. Finally, in both intervention groups that were compared, regularity (i.e. a fixed pattern of sleeping, waking up and feeding, playing together followed by playing alone at a fixed place and with the first signs of tiredness put into bed while awake) and regularity with swaddling added, the crying reduction was similar and independent of the emotional state of the mothers.

Introduction

Excessive crying in infants is a serious health problem. Crying for more than 3 hours a day for at least one day in the preceding week occurred in 13% and 8.6% respectively in two large Dutch prevalence studies in 1999[1] and 2003[2]. Various causes can be found for excessive crying, such as cow's milk allergy, gastro-oesophageal reflux, infections, or various rare conditions, but in most cases excessive crying remains unexplained.[3,4]
Several prospective studies described risks factors for having an excessively crying infant, such as anxiety in pregnancy, depressive symptoms during pregnancy, parenting stress, job strain during pregnancy, and high maternal scores for negative life changes.[5-8] Furthermore, very young women, women who were not living with the father, and women with high trait anxiety, measured during pregnancy, have infants who cry more at the age of 5 weeks.[9,10] Rautava showed prospectively that experience of stress and physical symptoms during pregnancy, dissatisfaction with the sexual relationship, and negative experiences during childbirth were associated with the development of excessive crying.[11] Retrospectively, a subjectively troubled pregnancy and a more negative experience of delivery by the mother seemed to predict a higher risk of infant crying.[12,13]

Methods

We conducted a randomised trial into the added effect of swaddling on excessive crying, comparing regularity (i.e. a fixed pattern of sleeping, waking up and feeding, playing together followed by playing alone at a fixed place and with the first signs of tiredness put into bed while awake) with regularity supplemented with swaddling.[2] During this study we collected data on events related to pregnancy. The psychological characteristics of parents who seek help and/or are referred for the excessive crying of their young infant will be described in this article, and compared to controls. Furthermore, we investigated whether the outcome (mean crying and cry perception) in the two intervention groups (regularity compared to regularity supplemented with swaddling) differed between mothers with a high score on mental health scales and those with a low or normal score.

Design – Randomised trial: Diet+ and I-group

A standardized case definition with the modified Wessel's criteria of excessive crying (i.e., crying for more than 3 hrs/24hr for at least 3 days a week) and parental perception of their infants' crying was used for recruitment.[14]
Inclusion criteria were healthy infants who cried excessively with a maximum age of 12 weeks and 6 days and a minimal gestational age of 32 weeks.

A baseline week was used to validate the crying by means of a 24-hour diary,[2,15] to collect extensive parental and psychological data and to assess the effect of a hypoallergenic diet (hypoallergenic formula or, in case of breastfeeding, a maternal diet free of cow's milk, egg, (shell) fish, and nuts). The group whose crying decreased to less than 2 hrs/24 hr from day three/four of the baseline onwards and/or whose parents did not perceive them as crying excessively anymore (DIET+ group), were excluded. Those infants that did not reduce their crying went on to the intervention study (I-group).

Design: control group

Control data for the initial interview and baseline measurements were obtained by means of written questionnaires, identical to the one filled out by parents included in the randomised trial (both DIET+ and I-group) and handed out at 14 midwifery practices shortly before the expected date of birth, with a self-addressed envelope (SAE) included. Parents were asked to fill in the questionnaire at a specified age between 1 and 12 weeks. The age distribution mirrored those of the excessively crying infants.

Measurements

Parents in the randomised trial recorded infant behaviour in a *24-hour diary*.[2,15] Furthermore, maternal perception of infant crying (eight 7-point Likert-type scales: urgent, grating, sick, arousing, piercing, discomforting, aversive, distressing, and manipulative) and the reaction to the cry (four 7-point Likert-type scales: anger/irritation, sadness, spoiled, care for) were recorded twice during the baseline week in the *Cry Perception Scale*.[16] From the baseline week diaries we calculated the total and mean duration of crying, fussing, sleeping, being awake alone and content, playing with the baby, feeding, caring and crying/fussing (min/24 hr).
At the moment of inclusion, both trial and control parents completed a questionnaire on medical and psychological measures on pregnancy, delivery and the postnatal period to assess their influence.[11,13,17]

Standardized instruments used were:

- *Pregnancy Related Anxiety Questionnaire – Revised (PRAQ-R)*[18]

The PRAQ-R is an abbreviated version of the PRAQ[19], which measures specific fears and worries related to pregnancy. This shortened 34-item version was derived from the original version by retaining the items with the highest factor loadings on each of the five subscales: 'fear of giving birth' (8 items), 'fear of bearing a physically or mentally handicapped child' (5 items), 'fear of changes and disillusion in partner relationship' (6 items), 'fear of changes' (4 items) and 'concern about one's mental well-being and the mother-child relationship' (4 items). Items were scored on a 5-

point scale (4 = very true, 3 = true, 2 = sometimes true, 1 = not true, 0 = definitely not true). A high score means more maternal anxiety and pregnancy related fears.

- *General Health Questionnaire 28 (GHQ-28)*[20]

The General Health Questionnaire (GHQ) is a self-administered screening test, designed to identify short-term changes in mental health (depression, anxiety, social dysfunction and somatic symptoms). The GHQ focuses on the client's ability to carry out 'normal' functions and the appearance of any new disturbing phenomena. The GHQ-28 is the most applied version of the GHQ. A high score on the scale is related to a more negative emotion.

- *Revised Profile of Mood States (POMS-R)*[21]

The POMS–R is a self-report, observer rated measure of mood states that can be used to monitor transient or fluctuating affective states in therapeutic and research environments. The POMS-R provides scores on six subscales: 'depression' (eight items, range 0-32), 'anger' (seven items, range 0-28), 'fatigue' (six items, range 0-24), 'vigor' (five items, range 0-30), and 'tension' (six items, range 0-24). All 32 items were recoded into the same direction and a low score reflects a more negative mood.

- *Social Support List – Discrepancies (SSL-D)*[22]

The SSL – D is a self-report, observer rated measure of the dissatisfaction with the received social support from the primary support group [parents (-in-law), brothers (in-law), and sisters (-in-law)]. The SSL – D provides scores on six subscales: 'daily emotional support' (four items), 'emotional support with problems' (eight items), 'appreciation of support' (six items), 'instrumental interaction' (seven items), 'social companionship' (five items), and 'informative support' (four items). All 34 items were recoded into the same direction (range of total score: 22-102). A high score on the scale reflects more dissatisfaction concerning the experienced social support.

For the SSL – D, the GHQ – 28 and the POMS – R we used mean score + 2 SD of our control mothers to define the clinical cut-off score for this group of mothers.

Statistical Analysis

All data were entered and analysed in Statistical Package for Social Science (SPSS) for Windows 12.0 (SPSS Inc., Chicago, Ill.). T-tests, Pearson χ^2 tests, Mann-Whitney U tests and ANOVA's were used to analyse the demographic data and the Cry Perception Scale. In multivariate analyses we corrected for age of the baby (wks), birthweight (grams), gender, birth by suction, age mother at first live birth (yrs), parental age at time of interview (yrs), maternal level of education, maternal current smoking behaviour and maternal smoking behaviour during pregnancy.

Results

Response

In the randomised trial, 398 infants were included and the intervention was completed by 382 infants (I-group, Figure 1).[2] After the baseline week, crying reduced to less than 2 hrs/24hr in 47 infants (9.5%) or parents felt that the problem of excessive crying had been solved, and so they were excluded before the intervention started (DIET+, Figure 1). From the total of 1330 self-addressed envelopes handed out to control parents at midwifery practices, 412 were returned (31%). Control parents assessed the amount their infant cried. If this was more than 3 hours a day, the infant was excluded from the control data. Of the controls 23 (5.6%) were excluded due to excessive crying, ten infants were older than 12 weeks (2.4%), 2 mothers had a pregnancy duration < 32 weeks (0.5%), and 10 questionnaires were incomplete (2.4%) (Figure 1).

Demographic characteristics

Demographic factors were determined for the I-group, DIET+ and controls, and compared to population data (Table 1). DIET+ mothers were younger at first live birth compared to the mothers in both other groups (Table 1) and they were less often university-educated. Control data are similar to the population data; only birthweight is higher in controls.

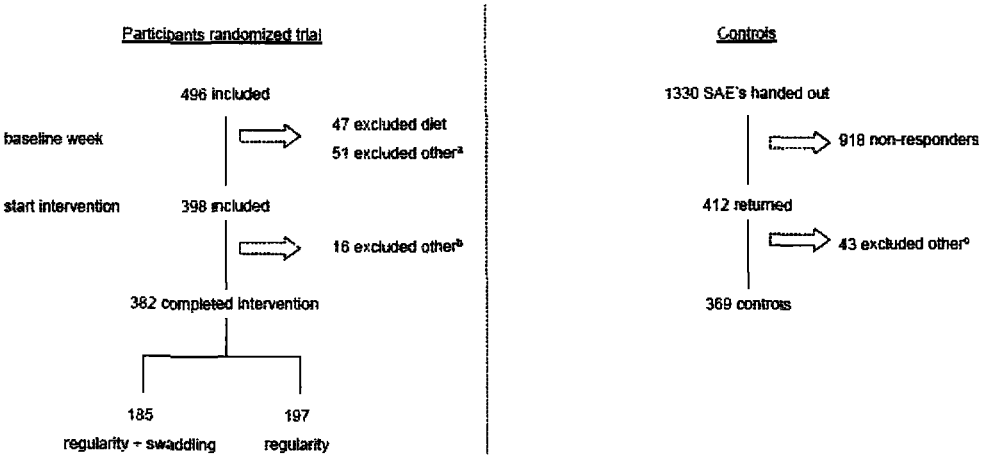


Figure 1. In- and excluded cases.

a. 2 not meeting inclusion criteria, 15 sudden unexplained reduction of crying, 6 admitted to hospital, 2 started swaddling, 7 could not identify one self with the given advice, 4 visited alternative health care, 2 treatment of thrush, 3 treatment of reflux, 1 treatment with lactulose, 7 reduction of crying because of malnutrition, 2 transport problem
b. 1 not meeting inclusion criteria, 1 admitted to hospital, 1 started swaddling, 12 could not identify one self with the given advice, 1 visited alternative health care
c. 23 cried excessively, 10 not meeting inclusion criteria, 10 incomplete questionnaires

Physical and psychological features during pregnancy, delivery and childbed

Compared to controls, both mothers of the I-group and DIET+ group reported a significantly higher incidence of abnormalities in their pregnancy (i.e. delayed infant growth, early contractions, twin or multiple pregnancy, abnormal loss of blood, high blood pressure, narrow pelvis, or reversed position of the baby) (adjusted OR 2.33, 95% CI 1.66 – 3.28, p 0.000 resp. adjusted OR 2.65, 95% CI 1.51 – 4.61, p 0.001) and reported a significantly higher incidence of illnesses they suffered from (diabetes, epilepsy, decreased or increased activity of the thyroid gland, high blood pressure, allergies, rheumatism, celiac disease, psychic complaints, or recurrent urinary tract infections) (adjusted OR 1.68, 95% CI 1.89 – 2.04, p 0.000 resp. adjusted OR 1.60, 95% CI 1.14 – 2.25, p 0.007). On both items, no significant differences between DIET+ mothers and I-group mothers were found (data not shown).
In the first trimester of pregnancy, mothers of the I-group were more afraid of pain during delivery (crude OR 1.39, 95% CI 1.01 – 1.91, p 0.046) compared to DIET+ mothers. This difference disappears, however, after correcting in the multivariate analyses. Compared to controls, I-group mothers were more afraid they would scream during delivery (adjusted OR 1.21, 95% CI 1.02 – 1.44, p 0.028), or that their infant would become a physically ill or weak child (crude OR 1.16, 95% CI 1.03 – 1.30, p 0.016), although this last difference disappears in the multivariate analyses. Comparing the DIET+ to the control mothers, no differences were found on the scores of the PRAQ–R (data not shown).

Table 1. Demographic characteristics for intervention infants, DIET+, and control infants compared to population data.

Parental features	I-group (n=382) N (%) / Mean ± SD	DIET+ (n=47) N (%) / Mean ± SD	Controls (n=369) N (%) / Mean ± SD	Population data (n=200.210)[I]
Age of mother at first live birth (years) *^#	29.81 ± 4.48	26.53 ± 3.87	28.22 ± 4.21	29.4
Age of mother (years) *^	32.06 ± 4.46	29.78 ± 4.95	30.98 ± 4.33	31.1
Maternal education *^				
- no education/primary school/lbo/mavo[1]	85 (24.8)	11 (26.9)	95 (27.5)	30.2 (%)
- havo/vwo/mbo[2]	128 (37.3)	22 (53.7)	165 (47.7)	32.5 (%)
- hbo/university[3]	130 (37.9)	8 (19.5)	86 (24.9)	37.3 (%)
Marital status *				
- living together with partner and infant(s)	363 (95.5)	44 (95.7)	352 (97.0)	97.9 (%)
Maternal smoking during pregnancy	61 (16.0)	11 (23.4)	52 (14.1)	13.3 (%)
Child features				
Gender - boy	201 (52.6)	25 (53.2)	190 (51.6)	102.870 (51.4)
Birth order - first born^	208 (54.5)	26 (55.3)	156 (42.4)	91.120 (45.5)
Age of infant at enrolment (weeks) ^#	7.98 ± 2.52	8.47 ± 2.43	7.17 ± 2.3	-
Birth weight (gram) ^#	3382.73 ± 579.75	3422.43 ± 612.29	3613.61 ± 524.65	3459
Pregnancy duration (weeks) ^	39.63 ± 1.64	39.61 ± 1.87	39.99 ± 1.34	40
Delivery duration (hours) *^	11.97 ± 12.6	7.51 ± 7.82	9.5 ± 10.64	-

1. Lower technical and vocational training and lower general secondary education
2. Intermediate vocational training and advanced secondary education
3. Higher vocational education (college education) and university

* Significant difference between I-group and DIET+

^ Significant difference between I-group and controls

\# Significant difference between DIET+ and controls

I. Schaijk M van, Lanting CI, Wouwe JP van, Engelberts AC, L'Hoir MP. Sample survey neonatal risk factors for cot death. November 2002 – April 2003 [In Dutch]. TNO Quality of Life: Leiden, 2006

According to the questionnaires developed by Rautava and Høgdall [11,13] and translated by us, mothers of the I-group felt to be on their own during pregnancy compared to DIET+ mothers (crude OR 1.54, 95% CI 1.11 – 2.12, p 0.010; adjusted OR 1.54, 95% CI 1.10 – 2.17, p 0.013). When comparing the I-group and DIET+ group with controls, on almost all of these items both groups experienced more stress (data not shown).

Mothers in the I-group contrary to mothers in the DIET+ group, reported having more physical complaints during pregnancy (i.e. fatigue, nausea, acidity, stabbing pain on the chest, puffy feeling, constipation, varicose veins, pelvic instability, and belly contractions) (adjusted OR 1.12, 95% CI 1.02 – 1.24, p 0.019) and more symptoms of illness during pregnancy (migraine, headache, backache, swollen hands and feet, and skin problems) when compared to controls (adjusted OR 1.25, 95% CI 1.07 – 1.45, p 0.004). On both items, no significant differences between DIET+ mothers and I-group mothers were found (data not shown). Both groups had an equal assumption of their own health during pregnancy (crude OR 1.02, 95% CI 0.90 – 1.16, p 0.729), but when compared to controls, I-group mother rated their health during pregnancy significantly more poorly (adjusted OR 1.40, 95% CI 1.24 – 1.57, p 0.000). DIET+ mothers were only significantly different in the univariate analysis, in the multivariate analysis the difference disappeared (crude OR 1.30, 95% CI 1.06 – 1.60, p 0.011; adjusted OR 1.21, 95% CI 0.99 – 1.51, p 0.094).
To examine the role of multiple risks, we summed the three individual prenatal risk factors: pregnancy related anxiety symptoms, physical complaints, and symptoms of illness during pregnancy, and we created an index that ranged from zero to three risk factors. Within the I-, DIET+ and control group mothers, 11 (1.4%) had zero risk factors, 196 (25.0%) had one risk factor, 534 (68.2%) had two, and 42 (5.4%) had three risk factors. When comparing I-group mothers and DIET+ mothers to controls, no differences were found in the amount of prenatal risk factors (data not shown).

Current psychological features

Several questions pertained to state of mind at inclusion in the randomised trial for infant crying, or for controls when filling out the questionnaire. Scores on subscales and total scores of the GHQ-28, POMS – R and SSL – D showed no significant differences between the I-group and the DIET+ group (data not shown). Compared to controls, mothers of the I-group and the DIET+ group both score significantly more negative on POMS – R and GHQ – 28 and all its subscales (Table 2,3). When looking at social support (SSL-D), I-group mothers experience significantly less social support on almost all items, while the DIET+ mothers only differ from controls on the item 'emotional support with problems' (Table 2,3).

Table 2. Maternal scores on psychosocial scales PRAQ - R, GHQ - 28, POMS - R, and SSL - D

	I-group (n=382)	DIET+ (n=47)	Controls n=369)
Pregnancy Related Anxiety Questionnaire-Revised (PRAQ - R)			
Fear of pain during delivery and childbirth	1.9 ± 1.30	1.5 ± 0.95	1.8 ± 1.18
Fear of giving birth	1.7 ± 1.17	1.5 ± 0.85	1.5 ± 1.05
Fear of screaming during delivery	1.6 ± 1.03	1.4 ± 1.00	1.4 ± 0.88
Fear of bearing a mentally handicapped child	2.7 ± 1.31	2.4 ± 1.16	2.6 ± 1.22
Fear that the infant would die just after childbirth	2.3 ± 1.31	2.0 ± 1.17	2.1 ± 1.16
Fear of bearing a physically handicapped child	2.8 ± 1.25	2.4 ± 1.21	2.6 ± 1.22
Fear of bearing a physically ill or weak child	2.5 ± 1.28	2.1 ± 1.16	2.3 ± 1.17
General Health Questionnaire - 28			
Total score (GHQ - 28)	33.6 ± 12.01	31.6 ± 11.06	19.2 ± 8.46
GHQ Somatic Symptoms (A)	9.6 ± 4.26	9.3 ± 4.37	5.9 ± 3.26
GHQ Fears and sleeplessness (B)	11.0 ± 4.62	10.1 ± 4.79	5.2 ± 4.05
GHQ Social Dysfunction (C)	10.9 ± 3.72	10.9 ± 2.78	7.6 ± 2.27
GHQ Grave Depression (D)	2.1 ± 3.32	1.3 ± 2.02	0.5 ± 1.31
Revised Profile Of Mood States (POMS - R)			
POMS-r Depression I	8.6 ± 7.1	7.3 ± 5.18	3.4 ± 5.81
POMS-r Anger II	7.3 ± 5.97	7.3 ± 5.59	4.4 ± 5.43
POMS-r Fatigue III	11.9 ± 6.23	11.0 ± 6.52	5.9 ± 5.15
POMS-r Vigor IV	11.5 ± 4.24	11.0 ± 3.92	8.0 ± 3.98
POMS-r Tension V	7.5 ± 5.30	6.9 ± 4.40	3.7 ± 4.54
Social Support List - Discrepancies			
Total score (possible range: 34-102) (SSL - D)	46.1 ± 12.44	44.3 ± 10.04	42.1 ± 8.83
SSL-D 'daily emotional support'	6.1 ± 2.33	5.5 ± 2.07	5.4 ± 1.81
SSL-D 'emotional support with problems'	11.4 ± 3.91	11.5 ± 3.85	9.6 ± 2.54
SSL-D 'appreciation of support'	7.7 ± 2.35	7.5 ± 1.91	7.4 ± 1.95
SSL-D 'instrumental interaction'	8.7 ± 2.30	8.2 ± 1.77	8.0 ± 1.62
SSL-D 'social companionship'	7.1 ± 2.57	6.6 ± 1.85	6.9 ± 2.19
SSL-D 'informative support'	5.1 ± 1.63	5.0 ± 1.13	4.6 ± 1.17

Compared to DIET+ mothers, I-group mothers felt to be left on their own in their current situation (crude OR 1.23, 95% CI 1.03 - 1.49, p 0.026; adjusted OR 1.26, 95% CI 1.03 - 1.54, p 0.023). They also experienced their infants as more vulnerable (crude OR 1.23, 95% CI 1.04 - 1.46, p 0.017; adjusted OR 1.24, 95% CI 1.03 - 1.50, p 0.022).
Compared to controls, mothers of excessive criers (I-group and DIET+) experienced their child as more vulnerable and difficult (data not shown).

Comparison of parents in the clinical range vs. non-clinical range

On all psychological questionnaires, the proportion of mothers of the I-group and the DIET+ group scoring in the clinical range (defined by the cut-off point 'mean controls + 2 SD') is significantly higher compared to the proportion of control mothers scoring in the clinical range (Table 4).
When selecting all parents within the I-group who are in the clinical range of the GHQ - 28, SSL - D or all subscales of the POMS - R, the reduction in crying (less or more than 25%) is equal in both intervention groups (regularity vs. regularity with

swaddling added) and independent of the age of the baby at inclusion. The same results are found for the parents within the I-group who are in the non-clinical range. Infants of mothers within the clinical range of the GHQ – 28 (total score and 'fear and sleeplessness' and 'social dysfunction') and POMS – R ('depression' and 'fatigue') cried significantly more at inclusion than infants whose mothers did not score in the clinical range. This difference disappears within all subscales after the first week, except for GHQ –28 'social dysfunction'. Infants of mothers who score in the clinical range of the GHQ – 28 'social dysfunction' cry significantly more during almost the total intervention period. Mothers who are in the clinical range of (sub-) scales of the GHQ – 28, the POMS – R, and the SSL – D, perceive their infants' crying as more negative (Cry Perception Scale), except for the items 'spoiled' and 'care for'. On these two items mothers in the non-clinical range score more negatively.

Age at inclusion and psychological features

Within the I-group, infants' age at inclusion is positively correlated with a more negative score on the depression scale of the GHQ – 28 (R 0.138, p .007), as well as higher scores (indicative of more dissatisfaction) on the SSL – D subscales 'daily emotional support' (R 0.104, p .045), 'instrumental interaction' (R 0.115, p .027), and 'informative support' (R 0.114, p .028). All other subscales of the GHQ – 28, the SSL – D, and the POMS – R do not correlate to age of the baby at inclusion. In the DIET+ group, a higher age of the baby at inclusion is negatively correlated with the POMS – R subscale 'fatigue' (R -.307, p 0.04).

Cry Perception Scale

At baseline, the DIET+ mothers perceived their infants' cry as significantly unhealthier (crude OR 1.24, 95% 1.02 – 1.51, p 0.030), although in the multivariate analysis this difference disappears (adjusted OR 1.19, 95% CI 0.97 – 1.47, p 0.093). None of the other items differed between the I- and the DIET+ group.
Compared to controls, I-group and DIET+ mothers score significantly more negative on all 'perception'-items (data not shown), and the crying of both groups elicited significantly more negative 'anger' (adjusted OR 2.14, 95% CI 1.87 – 2.44, p 0.000; resp. adjusted OR 2.02, 95% CI 1.56 – 2.63, p 0.000)) and 'sadness' (adjusted OR 2.70, 95% CI 2.33 – 3.13, p 0.000; resp. adjusted OR 3.85, 95% CI 2.60 – 5.68, p 0.000) responses, and, only in the I-group, a more negative 'care for' response (adjusted OR 0.85, 95% CI 0.76 – 0.95, p 0.004).

Table 3. Crude and adjusted* Odds Ratios for psychosocial scales GHQ – 28, POMS – R, and SSL – D

	I-group vs. DIET+		I-group vs. controls		DIET+ vs. controls	
	Crude OR (95% CI)	Adjusted* OR (CI 95%)	Crude OR (95% CI)	Adjusted* OR (95% CI)	Crude OR (95% CI)	Adjusted* OR (CI 95%)
General Health Questionnaire – 28						
Total score (GHQ – 28)	ns	ns	p 0.000	1.14 (1.11-1.16)	p 0.000	1.12 (1.09-1.18)
GHQ Somatic Symptoms (A)	ns	ns	p 0.000	1.30 (1.23-1.37)	p 0.000	1.28 (1.18-1.41)
GHQ Fears and sleeplessness (B)	ns	ns	p 0.000	1.30 (1.25-1.35)	p 0.000	1.27 (1.18-1.37)
GHQ Social Dysfunction (C)	ns	ns	p 0.000	1.47 (1.37-1.56)	p 0.000	1.69 (1.45-1.96)
GHQ Grave Depression (D)	ns	ns	p 0.000	1.45 (1.30-1.64)	p 0.000	1.35 (1.15-1.59)
Revised Profile Of Mood States (POMS – R)						
POMS-r Depression I	ns	ns	p 0.000	1.15 (1.11-1.18)	p 0.000	1.09 (1.04-1.14)
POMS-r Anger II	ns	ns	p 0.000	1.11 (1.08-1.14)	p 0.002	1.09 (1.03-1.14)
POMS-r Fatigue III	ns	ns	p 0.000	1.18 (1.14-1.20)	p 0.000	1.16 (1.10-1.22)
POMS-r Vigor IV	ns	ns	p 0.000	1.22 (1.16-1.27)	p 0.000	1.19 (1.10-1.30)
POMS-r Tension V	ns	ns	p 0.000	1.18 (1.14-1.22)	p 0.000	1.15 (1.06-1.19)
Social Support List – Discrepancies						
Total score (possible range: 34-102) (SSL – D)	ns	ns	p 0.000	1.03 (1.02-1.05)	ns	ns
SSL-D 'daily emotional support'	ns	ns	p 0.000	1.15 (1.06-1.25)	ns	ns
SSL-D 'emotional support with problems'	ns	ns	p 0.000	1.18 (1.11-1.23)	p 0.000	1.16 (1.06-1.28)
SSL-D 'appreciation of support'	ns	ns	ns	ns	ns	ns
SSL-D 'instrumental interaction'	ns	ns	p 0.000	1.20 (1.10-1.32)	ns	ns
SSL-D 'social companionship'	ns	ns	ns	ns	ns	ns
SSL-D 'informative support'	ns	ns	p 0.000	1.20 (1.08-1.35)	ns	ns

* corrected for age of the baby (wks), birth weight (grams), gender, birth by suction, age mother at first live birth (yrs), age mother at time of interview (yrs), age father at time of interview (yrs), maternal level of education, smoking during pregnancy, current smoking behaviour

Table 4. Prevalence rates of mothers of excessively crying infants scoring in the clinical range of the psychological scales GHQ – 28, POMS – R, and SSL – D

	I-group (n=382) N (%)	DIET+ (n=47) N (%)	Controls (n=369) N (%)
General Health Questionnaire – 28			
Total score ^#	146 (38.3)	12 (26.1)	18 (4.9)
GHQ Somatic Symptoms (A) ^#	91 (23.8)	15 (32.6)	17 (4.6)
GHQ Fears and sleeplessness (B) ^#	126 (33.1)	11 (23.9)	16 (4.4)
GHQ Social Dysfunction (C) ^#	124 (32.6)	14 (30.4)	11 (3.0)
GHQ Grave Depression (D) ^#	87 (22.8)	7 (15.2)	13 (3.6)
Revised Profile Of Mood States			
POMS-r Depression I ^	78 (20.5)	5 (11.1)	15 (4.1)
POMS-r Anger II *^	46 (12.1)	1 (2.2)	20 (5.5)
POMS-r Fatigue III ^#	120 (31.5)	9 (20.0)	16 (4.4)
POMS-r Vigor IV ^	50 (13.1)	3 (6.7)	9 (2.5)
POMS-r Tension V ^	62 (16.3)	4 (8.9)	14 (3.8)
Social Support List – Discrepancies			
Total score (possible range: 34-102)	54 (14.7)	7 (15.6)	20 (5.5)
SSL-D 'daily emotional support' ^	57 (14.9)	5 (10.6)	20 (5.4)
SSL-D 'emotional support with problems' ^#	84 (22.8)	9 (20.0)	26 (7.2)
SSL-D 'appreciation of support' ^	36 (9.8)	3 (6.7)	19 (5.3)
SSL-D 'instrumental interaction' ^	46 (12.5)	2 (4.4)	17 (4.7)
SSL-D 'social companionship' ^	37 (10.1)	1 (2.2)	18 (4.9)
SSL-D 'informative support' ^	39 (10.5)	2 (4.4)	16 (4.4)

* Significant difference between I-group and DIET+ (univariat)
^ Significant difference between I-group and controls (univariat)
\# Significant difference between DIET+ and controls (univariat)

Discussion

In a randomised trial we compared regularity with regularity supplemented with swaddling.[2] During this study, psychological factors possibly related to excessive crying were collected and compared to separate control data.

Weaknesses and strengths

The DIET+ infants reduced their crying after a week hypoallergenic diet. This does not prove that these infants all have a cow's milk allergy, although undoubtedly some have suffered from this. In part we expect that also this group includes infants that would have reduced their crying anyway, due to the natural course of infant crying. Furthermore, some families might react favourably to the extra attention given to them by the study itself. Nonetheless, it can be supposed that if infant crying is determined by unfavourable psychological conditions of the mother, the infants would not be in this group, and these DIET+ mothers would then resemble "normal" control mothers much more than the I-group.
To determine whether any predisposing child or maternal factors play a role in the onset of excessive crying, these factors should be assessed before the excessive crying has started, and ideally in pregnancy or at birth. Our questionnaires were filled out when the baby was several weeks old. The mood state at time will obviously colour the recollection of pregnancy and delivery. On the other hand, if a mother seeks professional help with an excessively crying infant, her actual mood is also of influence when answering questions on pregnancy and delivery. The answers are therefore clinically relevant.

Pregnancy

In a prospective community-based study, Van der Wal and colleagues showed that women who have depressive symptoms during pregnancy, pregnancy related anxiety, parenting stress, and job strain during pregnancy, are at risk for having an excessively crying infant.[5] Women who have multiple of these antenatal risk factors are at particular risk.[5] Rautava showed prospectively that experience of stress and physical symptoms during pregnancy, dissatisfaction with the sexual relationship, and negative experiences during childbirth were associated with the development of excessive crying in a baby.[11] In a study by Zuckerman, pregnant women who had a higher score on the Centre for Epidemiological Studies – Depression (CES – D) questionnaire for depressive symptoms were more likely to have an infant that would be inconsolable or cry excessively (aged 8-72 hours).[8] In a prospective study of Canivet, very young women, women who did not live with the father, and women with high trait anxiety were at a higher risk for having an infant that cried.[9] Søndergaard showed in a large prospective cohort study that mothers who reported distress during pregnancy had a threefold risk of excessive infant crying, and almost a twofold risk was found for the women who scored high on the psychological

distress scale, filled out during pregnancy (based on the Symptoms Check List (SLC – 90) and the GHQ).[7]
Miller found no difference in distress (GHQ – 28) in the third trimester in pregnant women related to infant crying/fussing behaviour at six weeks, the period at which crying is at its peak.[23] Høgdall found in a retrospective study that a subjectively troubled pregnancy was correlated with a higher risk of infant crying.[13]
In the present study, in which we divided mothers of infants who reduced their crying after a hypoallergenic diet from mothers of infants who did not do so, DIET+ mothers did not differ from control mothers on any item of the pregnancy related anxiety sub-scales. Mothers of the I-group, however, felt less supported by their partners during pregnancy, they experienced their childbed as more difficult and more often as a horrible time. These findings are derived from a non-validated questionnaire developed by Rautava and Høgdall [11,13] and translated by us, and should be interpreted cautiously. Our retrospective results are consistent with the prospective findings of Rautava, where parents who experienced stress or were socially isolated during pregnancy were more likely to have infants that cry excessively at the age of 3 months.[11]
Consistent with the prospective findings of Rautava[11] and the retrospective results of Høgdall[13], I-group mothers in our study, and not mothers in the DIET+ group, reported having more physical complaints and symptoms during pregnancy when compared to controls.

Current situation

Compared to DIET+ mothers, mothers of the I-group do not differ significantly in psychological features, as measured by the POMS – R, the GHQ – 28, and the SSL – D. The GHQ-28 is a pure state measure, responding to how much a subject feels that their present state "over the past few weeks" is different from their usual state. It does not make clinical diagnoses and can not be used to measure long-standing attributes.
The fact that mothers in the I-group of children that enrolled in the study at an older age scored higher on the GHQ-28 depression scale, suggests that depression is indeed a reaction to the situation. Increased maternal depressive symptoms however might also predispose for increased infant crying (based on baby day diaries), as two studies described, although a causal relationship between crying and maternal mood cannot be found.[23,24] The fact that mothers of the DIET+ group of older children at inclusion score higher on the item 'fatigue' of the POMS – R might be explained by the fact that these children started crying at a later age.
Compared to I-group mothers, mothers of DIET+ infants perceive their child's crying as unhealthier, which might either affirm a correct assessment of the cause of their infants crying or explain why they react more favourably to a diet as they would a priori be more likely to believe in its effect.
Compared to DIET+ mothers, mothers of the I-group emotionally experience more problems during their childbed. After delivery, mothers of the I-group had a higher

need for outside help, felt more limited and left on their own in their current situation, and they experienced their child as more vulnerable. Our retrospective results are again supported by Rautava's prospective.[11]
Within the I-group, some scales of the GHQ – 28 and the SSL – D, which indicate more dissatisfaction, correlate to age of the baby at inclusion. Only 'fatigue' (POMS – R) in the DIET+ group is related to age of the baby at inclusion. One might conclude that the longer the baby cries, the more negative feelings parents have, although a depression might also elicit the crying. A large Dutch population-based cohort study that included mothers in the 2nd trimester of pregnancy (the 'Generation-R Study') showed a significant relation between maternal depression, measured at 20-weeks pregnancy by means of the Brief Symptom Inventory (BSI)) and excessive crying – as perceived by the parents – at the postnatal infants age of 2 months.[25] Furthermore, paternal depression during pregnancy, measured with the same instrument, was significantly associated with excessive crying after birth.[25]
In a large prospective study, Canivet showed that a high trait anxiety, as measured in the 17th and again in the 35th-37th week of pregnancy, was associated with excessive crying after birth.[9] Younger and low educated mothers scored particularly high in trait anxiety. According to Akman, the mean Edinburgh Postpartum Depression Score (EPDS) of the mothers whose infants cried excessively according to Wessel's criteria (measured in the first week, first months and 4-6 months postpartum) was significantly higher than that of the mothers of infants who did not cry excessively.[26]

Clinical vs. non-clinical range

The proportion of mothers of the I-group and the DIET+ group scoring in the clinical range of all psychological measures is significantly higher compared to the proportion of mothers in the control group.
Within the I-group, more negative scores on the GHQ – 28 (total score and 'fear and sleeplessness', 'social dysfunction', and 'grave depression') and POMS – R ('depression', 'anger', 'fatigue', and 'tension') are associated with a higher amount of crying at inclusion. These correlations cannot be found within the DIET+ group and the association disappears within all subscales after the first intervention week, except for GHQ –28 'social dysfunction'. This is the only feature that predisposes a higher amount of crying during the following weeks. This could be an indication that social support is needed for families to perceive the study interventions as supportive. This does need further study.

In both intervention groups (regularity or regularity with swaddling added) excessive crying reduced equally in mothers of excessively crying infants whose prevalence rates of scores on GHQ – 28, POMS – R, and SSL – D are in the clinical range and mothers whose scores are not in the clinical range. This suggests that the supportive interventions are applicable for all mothers, with or without psychological problems.

Conclusion

In our study there are indications that mothers of excessively crying infants that do not respond to a week hypoallergenic diet differ from those that do, and from control mothers. Pregnancy and childbirth are viewed as more troublesome on several psychological and physical items. This finding is supported by previous studies showing that problems in pregnancy are related to excessive crying of the baby. When looking at psychological scores, the few differences between DIET+ and I- group mothers suggest that the higher proportion of clinical scores in both groups compared to controls is due to the excessive crying rather than a cause of the crying. Only on some items pertaining to social support I- group mothers score less favourably. It would seem logical to assume that adequate social support is an important factor that influences the perception and even the actual amount of crying. The outcome in both intervention groups, regularity and regularity with swaddling added, was not associated with psychological differences of the mothers.

References

1. Crying in babies. In: Brugman E, Reijneveld SA, Hollander-Gijsman ME den, Burgmeijer RJF, Radder JJ. Gauging in the child health care 1997-1998 (41-45). Leiden: TNO Prevention and Health, 1999
2. Sleuwen BE van, L'Hoir MP, Engelberts AC, Busschers WB, Westers P, Blom MA, Schulpen TWJ, Kuis W. Comparison of behaviour modification with and without swaddling as interventions for excessive crying. J Pediatr 2006;149:512-7
3. Nooitgedagt JE, Zwart P, Brand PLP. Causes, treatment and clinical outcome in infants admitted because of excessive crying to the paediatric department of the Isala clinics, Zwolle, the Netherlands, 1997/'03 [In Dutch]. Ned Tijdschr Geneeskd 2005;149:472-7
4. Lucassen PL, Assendelft WJ, Gubbels JW, van Eijk JT, van Geldrop WJ, Neven AK. Effectiveness of treatments for infantile colic: systematic review. BMJ 1998;316:1563-9
5. Wal MF van der, Eijsden M van, Bonsel GJ. Stress and emotional problems during pregnancy and excessive infant crying. J Dev Behav Pediatr 2007;28:431-7
6. Wurmser H, Rieger M, Domogalla C, Kahnt A, Buchwald J, Kowatsch M, Kuehnert N, Buske-Kirschbaum A, Papoušek M, Pirke K, von Voss H. Association between life stress during pregnancy and infant crying in the first six months postpartum: a prospective longitudinal study. Early Hum Dev 2006;82:341-9
7. Sondergaard C, Olsen J, Friis-Hasche E, Dirdal M, Thrane N, Sorensen HT. Psychosocial distress during pregnancy and the risk of infantile colic: a follow-up study. Acta Paediatrica 2003;92:811-6
8. Zuckermann B, Bauchner H, Parker S, Cabral H. Maternal depressive symptoms during pregnancy, and newborn irritability. J Dev Behav Pediatr 1990;11:190-4
9. Canivet CA, Östergren PO, Rosén A, Jakobsson I, Hagander BM. Infantile colic and the role of trait anxiety during pregnancy in relation to psychosocial and socioeconomic factors. Scan J Public Health 2005;33:26-34
10. Canivet CA, Östergren PO, Jakobsson I, Hagander BM. Higher risk of colic in infants of nonmanual employee mothers with a demanding work situation in pregnancy. Int J Behav Med 2004;11:37-47
11. Rautava P, Helenius H, Lehtonen L. Psychosocial predisposing factors for infantile colic. BMJ 1993;307:600-4
12. Räihä H, Lehtonen L, Huhtala V, Saleva K, Korvenranta H. Excessively crying infant in the family: mother-infant, father-infant and mother-father interaction. Child Care Health Dev 2002;28:419-29
13. Høgdall CK, Vestermark V, Birch M, Plenov G, Toftager-Larsen K. The significance of pregnancy, delivery and postpartum factors for the development of infantile colic. J Perin Med 1991;19:251-7

14. Wessel, MA, Cobb JC, Jackson EB, Harris GS, Detwiler AC. Paroxysmal fussing in infancy, sometimes called "colic". Pediatrics 1954;94:322-32
15. Barr RG, Kramer MS, Boisjoy C, McVey-White L, Pless IB. Parental diary of infant cry and fuss behaviour. Arch Dis Child 1988;63:380-7
16. Lester BM, Boukydis CFZ, Garcia-Coll CT, Holl WT, Peucker M. Infantile colic: cry characteristics, maternal perception of cry, and temperament. Infant Behav Dev 1992;15:15-26
17. Lucassen LB, Assendelft WJ, Gubbels LW, Eijk JT van, Douwes AC. Infantile colic: crying time reduction with a whey hydrolysate; a double blind, randomized placebo-controlled trial. Pediatrics 2000;106:1349-54
18. Huizink AC, Mulder EJH, Robles de Medina PG, Visser GHA, Buitelaar JK. Is pregnancy anxiety a distinctive syndrome? Early Hum Dev 2004;79:81-91
19. Van den Bergh B. The influence of maternal emotions during pregnancy on fetal and neonatal behavior. Pre-Peri-Nat Psych J 1990;5:119-30
20. Koeter MWJ, Ormel J. General Health Questionnaire [Dutch Translation]. Lisse: Swets & Zeitlinger BV, 1991
21. Ark, LA van der, Marburger, D, Mellenbergh GJ, Vorst HCM, Wald F. Revised Profile Of Mood States (POMS – r). Manual and Justification [In Dutch]. Nijmegen: Berkhout Nijmegen BV, 1995
22. Sonderen E van. Social Support List – Interactions (SSL – I) and Social Support List – Discrepancy (SSL – D). Manual [In Dutch]. University of Groningen, 1993
23. Miller AR, Barr RG, Eaton WO. Crying and motor behavior of six-week-old infants and postpartum maternal mood. Pediatrics 1993;92:551-8
24. Milgrom J, Westley DT, McCloud PI. Do infants of depressed mothers cry more than other infants? J Paediatr Child Health 1995;31:218-21
25. Berg MP van den. Parental psychopathology and the early developing child. The Generation R Study [Thesis]. Erasmus University Rotterdam, 2006
26. Akman I, Kusçu K, Özdemir N, Yurdakul Z, Solakoglu M, Orhan L, Karabekiroglu A, Özek E. Mothers' postpartum psychological adjustment and infantile colic. Arch Dis Child 2006;91:417-9

Do mothers of excessively crying infants differ psychologically from mothers of moderately crying infants?

7

Bregje E. van Sleuwen
Adèle C. Engelberts
Tom W.J. Schulpen
Wietse Kuis
Monique P. L'Hoir

Submitted for publication

Abstract

Objective
We studied maternal psychological characteristics in excessively or moderately crying infants, and in mothers who did not perceive their baby's crying as a problem. Furthermore, we compared two behavioural interventions, with and without swaddling in infants who cried excessively or moderately: a. regularity (i.e. a fixed pattern of sleeping, waking up and feeding, playing together followed by playing alone on a fixed place and at the first signs of tiredness put into bed while awake) and b. regularity with swaddling added.

Patients and Methods
In a randomised trial, 382 excessively crying infants completed a 3-month intervention, which offered either regularity or regularity with swaddling added. A control group of 368 infants who did not cry unduly was included.

Results
Mothers of excessively crying infants did not differ in pregnancy related anxieties compared to mothers of moderately crying infants or compared to control mothers, although mothers of moderate criers were more anxious than control mothers. On the Cry Perception Scale, mothers of excessive criers experienced the crying as more worrisome than mothers of moderate criers and control mothers. Mothers of moderately crying infants perceived crying significantly more often as 'spoiled' compared to controls. Mothers of excessively crying infants score more negative on the POMS – R 'depression' scale than mothers of moderate criers, but the two groups do not differ in scores within the clinical range. In both intervention groups (regularity vs. regularity with swaddling added) crying reduced equally and no differences were found between mothers of excessive and moderate criers.

Conclusion
Parents of excessively crying infants perceive their infant's cry behaviour as difficult, but this does not disturb their balance completely. Furthermore, psychological disturbances do not seem to be the cause, but rather the effect of the amount of crying, as only the item 'depression' shows a more negative score, but not on anxiety. The differences between mothers of excessive and moderate criers are relatively small, but mothers with moderate criers perceive it as more disturbing than mothers with infants that really cry excessively. As the two interventions (regularity vs. regularity with swaddling added) do not differ in outcome between mothers of excessively and moderately crying infants, this suggests that both interventions are applicable for the total range of mothers whose baby cries, regardless of the amount of crying.

Introduction

Excessive crying of infants is a serious health problem. In the Netherlands 22% of parents consult a well baby clinic doctor for excessive crying of their infant, 5% their General Practitioner and 2% the pediatrician.[1]

Excessive crying, defined as crying for at least 3 hours a day for one day or more in the preceding week, occurred in 13% and 8.6% respectively in two large Dutch prevalence studies in 1999 and 2003.[1,2] Wessel's definition of excessive crying[3], namely at least 3 hours on at least 3 days of the 3 previous weeks, occurs in 2-2.5% of the infant population.[1,2,4] In 1995, this definition was broadened to the 'modified Wessel's criteria' (crying for at least three hours a day three or more days a week).[5]

The perception of crying may differ from person to person, as well the stress it causes. To measure these subjective feelings, Lester developed the 'Cry Perception Scale'.[6] To quantify infant's crying, Barr developed and validated a 'Parental diary of cry and fuss behaviour'.[7] Crying can be seen as a parental complaint, in which parents decide if the amount of crying is excessive, or as an objectively measurable phenomenon, which either does or does not fulfil Wessel's criteria.[8] The group of infants that cry too much according to their parents can be divided into a group of moderately crying infants (< 3hrs/24hr) and a group of excessively crying infants (≥ 3hrs/24hr).[9-12] When looking at differences between these two groups, Räihä et al. found that parents of excessively crying infants and controls have stronger parental coalition and a better organised family structure when compared to parents of moderately crying infants, but both parents of moderate as well as excessive criers felt less close to each other and were less flexible compared to controls.[11] Parents of excessively crying infants had decreased ability to cope with daily activities, expressed the most unresolved problems, were less satisfied with each other, and showed less energy and vitality than parents of moderate criers and control parents.[11] In another case-control study, Räihä et al. described that fathers, and not mothers, of excessively crying infants were less expressive and had less warmth in their voice.[9] Furthermore, fathers of the excessive criers expressed less positive affect, they tended to be more anxious, they expressed less enthusiasm/joy of life, and they had less visual contact with their infant. Mothers of excessive as well as moderate criers showed 'meaningful communication' less frequently compared to controls, but more mothers were well connected/involved with their child. Mothers of moderate criers were less flexible in interaction and both parents were less creative/resourceful in both moderate and excessive criers. Parents of excessive criers showed weak coalition, lack of mutual closeness, and less efficient communication compared to control parents.[9]

In contrast, Papoušek and Von Hofacker found no differences between parents of moderate and excessive criers when compared to controls; mothers in both groups scored similarly low on feelings of self-efficacy, and high on depression, anxiety, exhaustion, anger, adverse childhood memories, and marital distress.[10] They suggested that these differences did not cause persistent crying, but perhaps maintained or exacerbated the problem.

We conducted a randomised trial comparing regularity (i.e. a fixed pattern of sleeping, waking up and feeding, playing together followed by playing alone on a fixed place and at the first signs of tiredness put into bed while awake) to regularity combined with swaddling.[2] After one intervention week, crying reduced in both groups with 42%, after two weeks with 50% and after 8 weeks with 75%. Regularity combined with swaddling reduced crying more only in infants aged 2-6 weeks, while offering regularity alone reduced crying more in the group of 7-12 weeks old infants. The group differences within each age category were approximately 10 minutes.
During this study we collected data on pregnancy-related events and psychological characteristics of parents who seek help and/or are referred for the excessive crying of their infant. We examined them in relation to the amount of crying by comparing the psychological profiles of mothers of moderately crying infants with those of mothers of excessively crying infants. Furthermore, we compared these with control data of mothers who did not perceive their infant as crying too much.
We also investigated whether mothers of excessively crying infants scored in the clinical range of the mental health scales more often compared to mothers of moderate criers. Finally, we investigated whether the effect of the two interventions (regularity compared to regularity combined with swaddling) is similar in the excessive criers compared to the moderate criers.

Methods

Design - cases

Cases were ascertained for a randomised trial on the effect of swaddling on excessive crying. A standardized case definition with the most commonly used modified Wessel's criteria of excessive crying (i.e., crying for at least 3 hrs/24hr for at least 3 days a week) was used for recruitment.[3,5]
Inclusion criteria were healthy infants with a maximum age of 12 weeks and 6 days and a minimal gestational age of 32 weeks who cried excessively according to their parents or healthcare workers. The baseline week was used to validate the crying,[7] collect parental data and to assess the effect of a hypoallergenic diet.[2] The group who ceased crying after hypoallergenic diet is excluded from the analyses.

Design

In the baseline week all infants received hypoallergenic diet (hypoallergenic formula or, in case of breastfeeding, a maternal diet free of cow's milk, egg, (shell) fish, and nuts). All infants whose crying had been reduced to less than 2 hrs/24 hr from day three/four of the baseline onwards and/or the parents, who perceived the crying as normalized, were excluded from the intervention study.
Control data for the initial interview and baseline measurements were obtained by means of written questionnaires, identical to the one filled out by parents included in

the randomised trial and handed out at 14 midwifery practices shortly before the expected date of birth, with a self-addressed envelope (SAE) included. Parents were asked to fill in the questionnaire at a specified age between 1 and 12 weeks. The age distribution mirrored those of the excessively crying infants.

Measurements obtained

Parents in the randomised trial recorded infant behaviour in a *24-hour diary*.[2,7] Furthermore, maternal perception of infant crying (eight 7-point Likert-type scales: urgent, grating, sick, arousing, piercing, discomforting, aversive, distressing, and manipulative) and the reaction to the cry (four 7-point Likert-type scales: anger/irritation, sadness, spoiled, care for) were recorded twice during the baseline week in the *Cry Perception Scale*.[6] A high score correlates with a more negative perception of the infants' crying.
At the moment of inclusion, both trial and control parents completed a questionnaire on medical and psychological measures on pregnancy, delivery and the postnatal period to assess their influence (Appendix A).[13-15]

Standardized instruments used were:

- *Pregnancy Related Anxiety Questionnaire – Revised (PRAQ-R)*[16]

The PRAQ-R is an abbreviated version of the PRAQ[17], which measures specific fears and worries related to pregnancy. A high score means more maternal anxiety and pregnancy related fears.

- *General Health Questionnaire 28 (GHQ-28)*[18]

The General Health Questionnaire (GHQ) is a self-administered screening test, designed to identify short-term changes in mental health (depression, anxiety, social dysfunction and somatic symptoms). The GHQ focuses on the client's ability to carry out 'normal' functions and the appearance of any new disturbing phenomena. A high score on the scale is related to more negative emotions.

- *Revised Profile of Mood States (POMS-R)*[19]

The POMS–R is a self-report, observer rated measure of mood states that can be used to monitor transient or fluctuating affective states in therapeutic and research environments. All 32 items were recoded into the same direction and a low score reflects a more negative mood.

- *Social Support List – Discrepancies (SSL-D)*[20]

The SSL – D is a self-report, observer rated measure of the dissatisfaction with the received social support from the primary support group [parents (-in-law), brothers (in-law), and sisters (-in-law)]. All 34 items were recoded into the same direction (range of total score: 22-102). A high score on the scale reflects more dissatisfaction regarding the social support experienced.

For the SSL – D, the GHQ – 28 and the POMS – R we used mean score + 2 SD of our control mothers to define the clinical cut-off score for this group of mothers.

Statistical Analysis

Data were entered and analysed in Statistical Package for Social Science (SPSS) for Windows 12.0 (SPSS Inc., Chicago, Ill.). T-tests, Pearson χ^2 tests, Mann-Whitney U tests and ANOVA's were used to analyse the demographic data, the Cry Perception Scale and the psychological measures.
In multivariate analyses we corrected for age of the baby (wk), birth weight (g), gender, birth by suction, age mother at first live birth (y), maternal and paternal age at time of interview (y), maternal level of education, smoking during pregnancy, and current smoking behaviour.

Cry features

From the baseline week diaries we calculated the total and mean duration of crying, fussing, sleeping, being awake alone and content, playing with the baby, feeding, caring and crying/fussing (min/24 hr).[2] Excessive crying was defined as crying ≥ 180 min/24hr, moderate crying was defined as crying < 180 min/24hr.

Results

Response

In the randomised trial 398 infants were included and the intervention was completed by 382 infants (Intervention group, I-group).[2] After the baseline week, crying reduced to less than 2 hrs/24hr in 47 infants (9.5%) and they were excluded before the intervention started according to protocol. From the total of 1330 self-addressed envelopes handed out to control parents at midwifery practices, 412 were returned (31%) (controls). Control parents assessed the amount their infant cried. If this was more than 3 hours a day, the infant was excluded from the control data. Of the controls 23 (5.6%) were excluded due to excessive crying, ten infants were older than 12 weeks (2.4%), 2 mothers had a pregnancy duration < 32 weeks (0.5%) and 10 questionnaires were incomplete (2.4%) (Figure 1).

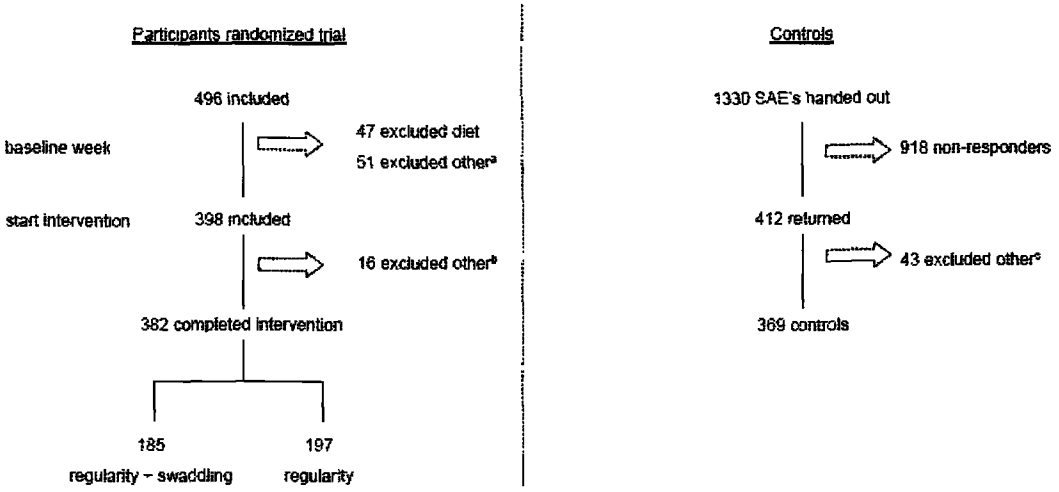


Figure 1. In- and excluded cases.

a. 2 not meeting inclusion criteria, 15 sudden unexplained reduction of crying, 6 admitted to hospital, 2 started swaddling, 7 could not identify one self with the given advice, 4 visited alternative health care, 2 treatment of thrush, 3 treatment of reflux, 1 treatment with lactulose, 7 reduction of crying because of malnutrition, 2 transport problem
b. 1 not meeting inclusion criteria, 1 admitted to hospital, 1 started swaddling, 12 could not identify one self with the given advice, 1 visited alternative health care
c. 23 cried excessively, 10 not meeting inclusion criteria, 10 incomplete questionnaires

Demographic characteristics

Demographic factors were determined for the I-group and control group. These last were compared to population data (Table 1). The two differences observed, birth weight and age, are controlled for in the multivariate analyses.

Table 1. Demographic characteristics for intervention and control infants compared to population data.

Parental features	Total group	I-group (n=382) Moderate criers	Excessive criers	DIET+ (n=47)	Controls (n=369)	Population data (n=200.210)[I,II]
Age of mother at first live birth (years)[b *^#]	29.81 ± 4.48	30.04 ± 4.43	29.40 ± 4.80	26.53 ± 3.87	28.22 ± 4.21	29.4
Age of mother (years)[b *^]	32.06 ± 4.46	32.34 ± 4.43	31.58 ± 4.83	29.78 ± 4.95	30.98 ± 4.33	31.1
Maternal education[a *^]						
- no education/primary school/lbo/mavo[1]	85 (24.8)	52 (21.1)	24 (23.5)	11 (26.9)	95 (27.5)	30.2 (%)
- havo/vwo/mbo[2]	128 (37.3)	90 (36.6)	41 (40.2)	22 (53.7)	165 (47.7)	32.5 (%)
- hbo/university[3]	130 (37.9)	104 (42.3)	36. (37)	8 (19.5)	86 (24.9)	37.3 (%)
Marital status[a *]						
- living together with partner and infant(s)	363 (95.5)	235 (96.3)	94 (92.2)	44 (95.7)	352 (97.0)	97.9 (%)
Maternal smoking during pregnancy[a]	61 (16.0)	35 (14.3)	19 (18.6)	11 (23.4)	52 (14.1)	13.3 (%)
Child features						
Gender[a] – boy[@]	201 (52.6)	120 (48.8)	63 (61.8)	25 (53.2)	190 (51.6)	102.870 (51.4)
Birth order[a^] - first born^	208 (54.5)	138 (36.1)	55 (53.9)	26 (55.3)	156 (42.4)	91.120 (45.5)
Age of infant at enrolment (weeks)[b ^#@]	7.98 ± 2.52	8.30 ± 2.53	7.31 ± 2.28	8.47 ± 2.43	7.17 ± 2.3	-
Birth weight (gram)[b ^#]	3382.73 ± 579.75	3398.61 ± 574.82	3371.86 ± 620.44	3422.43 ± 612.29	3613.61 ± 524.65	3459
Pregnancy duration (weeks)[b ^]	39.63 ± 1.64	39.51 ± 1.87	39.45 ± 1.82	39.61 ± 1.87	39.99 ± 1.34	40
Delivery duration (hours)[b *^]	11.97 ± 12.6	12.04 ± 12.34	12.46 ± 13.87	7.51 ± 7.82	9.5 ± 10.64	-
Amount of crying / 24hr		-	-			
- moderate (<3hrs/24hr)	246 (70.7)			30 (73.2)	-	-
- excessive (>3hrs/24hr)	102 (29.3)			11 (26.8)	-	-

a. counts (percentages)
b. mean ± SD

1. Lower technical and vocational training and lower general secondary education
2. Intermediate vocational training and advanced secondary education
3. Higher vocational education (college education) and university

^ Significant difference between I-group and controls
@ Significant difference between moderate and excessive criers

I. Schaijk M van, Lanting CI, Wouwe JP van, Engelberts AC, L'Hoir MP. Sample survey neonatal risk factors for cot death. November 2002 – April 2003 [In Dutch]. TNO Quality of Life: Leiden, 2006
II. Central Bureau for Statistics (CBS). Birth, height and weight at birth and breastfeeding rates [*In Dutch*]. Voorburg, 2006

Cry perception scale

Comparison of mothers of excessive criers with mothers of moderate criers shows that the latter score significantly less negatively on the maternal cry perception-items 'urging' (adjusted OR1.365, 95% CI 1.070 - 1.741, p 0.012), 'grating' (adjusted OR 1.438, 95% CI 1.086 - 1.905, p 0.011), 'sick' (adjusted OR 1.301, 95% CI 1.119 - 1.511, p 0.001), 'piercing' (adjusted OR 1.435, 95% CI 1.063 - 1.97, p 0.018) and 'distressing' (adjusted OR 1.239, 95% CI 1.043 - 1.472, p 0.015) and on the maternal cry reaction-item 'sadness' (adjusted OR 1.241, 95% CI 1.033 - 1.490, p 0.021) (Cry Perception Scale).[6] No other items differed significantly between the two groups. When comparing mothers of excessive criers with control mothers, the excessive group scored significantly more negative on all items of both scales, except for the 'spoiled' item (adjusted OR 1.160, 95% CI 0.981 - 1.372, p 0.083). Mothers of moderate criers showed the same pattern but on the item 'spoiled' (adjusted OR 1.209, 95% CI 1.064 - 1.373, p 0.004) they had a significantly more negative score than control mothers.

Psychological features during pregnancy, delivery and childbed

In the first trimester of pregnancy, mothers of excessively crying infants did not differ from mothers of moderately crying infants on any of the items of the PRAQ - R. Mothers of excessively crying infants also do not score differently on any of the PRAQ - R scales compared to controls. However, mothers of moderate criers score significantly higher on almost all items of the PRAQ - R compared to control mothers (Table 2).

Compared to mothers of moderate criers, mothers of excessive criers experienced their pregnancy more negatively (adjusted OR 1.461, 95% CI 1.166-1.830, p 0.001), they were more disappointed with their pregnancy (adjusted OR 1.340, 95% CI 1.111 - 1.617, p 0.002), and felt it was more difficult than expected (adjusted OR 1.295, 95% CI 1.053 - 1.592, p 0.014).

Compared to mothers of moderately crying infants, mothers of excessive criers felt that their infants cried a lot compared to other babies (adjusted OR 1.697, 95% CI 1.261 - 2.283, p 0.000), they could comfort their baby less easy (adjusted OR 1.511, 95% CI 1.142 - 1.996, p 0.004), they experienced the crying as a much bigger problem (adjusted OR 1.335, 95% CI 1.059 - 1.684, p 0.015) and they experienced their baby as less easy compared to other baby's (adjusted OR 1.395, 95% CI 1.110 - 1.751, p 0.004).

Table 2. Pregnancy Related Anxiety scores (revised) in the first trimester of pregnancy.

Parental features	I-group Excessive criers Mean ± SD	I-group Moderate criers Mean ± SD	Controls
Pregnancy Related Anxiety Questionnaire-Revised (PRAQ – R)			
Fear of pain during delivery and childbirth	1.96 ± 1.33	1.94 ± 1.31	1.80 ± 1.18
Fear of giving birth	1.64 ± 1.14	1.69 ± 1.20	1.51 ± 1.05
Fear of screaming during delivery ^	1.51 ± 0.95	1.58 ± 1.07	1.40 ± 0.88
Fear of bearing a mentally handicapped child ^	2.57 ± 1.41	2.76 ± 1.28	2.55 ± 1.22
Fear that the infant would die just after childbirth ^	2.33 ± 1.33	2.36 ± 1.29	2.14 ± 1.16
Fear of bearing a physically handicapped child ^	2.66 ± 1.34	2.80 ± 1.21	2.58 ± 1.22
Fear of bearing a physically ill or weak child ^	2.43 ± 1.35	2.52 ± 1.26	2.27 ± 1.17

^ Significant difference between I-group moderate and controls (univariate)

GHQ-28, POMS – R, SSL-D

Mothers of excessively crying infants did not score significantly higher on all (sub)scales of the SSL – D and GHQ – 28 compared to mothers of moderate criers, but after correcting for infants' age and gender, there is a small, but significant difference in the total score of the GHQ – 28 (adjusted OR 1.021, 95% CI 1.001 – 1.042, p 0.044).
When compared to moderate criers, mothers of excessively crying infants only score significantly higher on POMS – R 'depression' (adjusted OR 1.046, 95% CI 1.012 – 1.082, p 0.007).
Both mothers of excessively and moderately crying infants have significantly higher scores on the items of the GHQ – 28, POMS – R (sub) scales, and on the total and four subscales of the SSL – D when compared to controls (data not shown).

Scores of mothers in the clinical range

The same amount of mothers of moderately as of excessively crying infants score in the clinical range of all (sub)scales of the GHQ-28, POMS – R, and the SSL-D (Table 3). Compared to mothers of controls, mothers of both groups have significantly more negative scores on almost all (sub)scales, except for the SSL – D item 'appreciation of support', where the mothers of moderate criers score in the clinical range as often as control mothers.

Table 3. Amount of mothers scoring in the clinical range of psychosocial scales.

	I-group		Controls
Parental features	Excessive criers N (%)	Moderate criers N (%)	N (%)
General Health Questionnaire – 28 (GHQ – 28)			
Total score *^	45 (44.1)	94 (38.4)	18 (4.9)
GHQ Somatic Symptoms (A) *^	28 (27.5)	57 (23.2)	17 (4.6)
GHQ Fears and sleeplessness (B) *^	37 (36.3)	79 (32.1)	16 (4.4)
GHQ Social Dysfunction (C) *^	39 (38.2)	74 (30.3)	11 (3.0)
GHQ Grave Depression (D) *^	27 (26.5)	55 (22.4)	13 (3.6)
Revised Profile Of Mood States (POMS – r)			
POMS-r Depression I *^	28 (27.5)	46 (18.9)	15 (4.1)
POMS-r Anger II *^	17 (16.7)	26 (10.6)	20 (5.5)
POMS-r Fatigue III *^	40 (39.2)	73 (29.8)	16 (4.4)
POMS-r Vigor IV *^	15 (14.7)	30 (12.2)	9 (2.5)
POMS-r Tension V *^	20 (19.6)	37 (15.1)	14 (3.8)
Social Support List – Discrepancies			
Total score (SSL – D; possible range: 34-102) *^	17 (17.3)	25 (14.7)	20 (5.5)
SSL-D 'daily emotional support' *^	17 (16.7)	34 (13.8)	20 (5.4)
SSL-D 'emotional support with problems' *^	24 (24.5)	55 (23.1)	26 (7.2)
SSL-D 'appreciation of support' *	10 (10.3)	25 (10.4)	19 (5.3)
SSL-D 'instrumental interaction' *^	14 (14.3)	31 (13.1)	17 (4.7)
SSL-D 'social companionship' *^	11 (11.2)	22 (9.3)	18 (4.9)
SSL-D 'informative support' *^	12 (12.2)	25 (10.4)	16 (4.4)

Significant difference within I-group between mothers of moderate and excessive criers
* Significant difference within I-group between mothers of excessive criers and controls
^ Significant difference within I-group between mothers of moderate criers and controls

The amount of crying and group comparison

In both intervention groups (regularity (R) vs. regularity combined with swaddling (R+S)) the crying reduces equally in excessively crying infants (cry reduction R 37.7%, R+S 45.6%) and in moderate criers (cry reduction R 30.8%, R+S 33.8%) in the first intervention week (crude OR 1.003, 95% CI 0.995 - 1.011, p 0.473). The same is found for the total intervention period of 12 weeks (cry reduction moderate criers: R 66.2%, R+S 62.3%; cry reduction excessive criers: R 74.3%, R+S 83.6%; crude OR 0.999, 95% CI 0.993-1.004, p 0.604). When looking at the 8.6% (n=22) of the infants in which the crying did not reduce, there was no influence of the type of intervention when comparing excessive criers (n = 4) with moderate criers (n = 18) (crude OR 0.212, 95% CI 0.018 – 2.467, p 0.215).

Age at inclusion and psychological features

Infants' age at inclusion in the moderately crying group is positively correlated with a more negative score on de 'depression' scale of the GHQ - 28 (R .172, p 0.007), as well as a more negative mood score on the POMS – R subscale 'fatigue' (R .129, p 0.044), and the total score of the SSL – D (R .199, p 0.002) and all subscales (indicative of more dissatisfaction, data not shown).
No correlation is found between infants' age at enrolment and maternal scores on psychological scales in the excessively crying groups.

Fathers and parental relationship

Compared to partners of mothers of moderately crying infants, partners of mothers of infants who cry excessively experienced their wives pregnancy, as assessed by the mothers, as more negative (adjusted OR 1.312, 95% CI 1.038 – 1.658, p 0.023). On all other items concerning the partners, there is no difference between the moderate and excessive group (data not shown).

Discussion

In a randomised trial into the added effect of swaddling on excessive crying maternal psychological factors were collected and compared to control data.

Weaknesses and strengths

One of the strengths of this study is the amount of children included and the relatively small drop out rate in the randomised controlled trial. A weakness of this study is the relatively low response of the control group. We were not able to find out whether the group of controls who did not return the questionnaire differs from those who participated. Comparison with national data however, shows only a difference in infants' birth weight, which was taken into account in the multivariate analyses. In the Netherlands, a country with 185.057 newborns in 2006, 31.6% of the mothers delivered their child at home under supervision of a midwife and research is not as broadly embedded in these practices as it is in hospitals, which could explain part of the relatively low response.[21]

General discussion

To determine whether any predisposing child or maternal factors play a role in the onset of excessive crying, these factors should be assessed before the crying starts and ideally in pregnancy or at birth. Our questionnaires were filled out when the baby was several weeks old. The mood state at time will obviously colour the recollection of pregnancy and delivery. On the other hand, if a mother seeks professional help

with a crying infant, her actual mood is also of influence when answering questions on pregnancy and delivery. The answers are therefore clinically relevant.
The GHQ-28 is a pure state measure, responding to how much a subject feels that their present state "over the past few weeks" is unlike their usual state. It does not make clinical diagnoses and should not be used to measure long-standing attributes.
Mothers of infants who cry excessively do not differ from controls in their pregnancy related anxieties (PRAQ – R). In contrast, mothers of moderate criers score significantly higher on almost all items of the PRAQ – R compared to controls mothers. This suggests that mothers who ask help for perceived, but in fact not actually excessive amounts of crying, (which in fact encompasses the moderately crying group in this study), experience the less than 3 hours a day of crying as untenable, but also experienced more pregnancy-related fears. Mothers of excessively crying infants only score more negative on the POMS – R 'depression' compared to the moderate criers. Mothers of excessively crying infants compared to mothers of moderate criers experienced the crying of their infants as more devastating, but compared to controls as less spoiled. Mothers of moderate criers perceived crying, on the contrary, more often as spoiled compared to controls.
Compared to mothers of excessively crying infants, there is no difference in the amount of mothers of moderate criers scoring in the clinical range on all psychological measures. When compared to controls, the amount of mothers scoring in the clinical range is larger in mothers of moderate as well as in mothers of excessive criers on all (sub)scales.
These findings are partly in agreement with the study of Pauli-Pott, were mothers who complain about crying but who's infant's crying actually does not meet Wessel's criteria, described significantly higher feelings of rejection and negative affect in response to their infant's crying, both compared to mothers who complained and did in fact have an excessively crying child as well as compared to control mothers who did not complain about the crying of their infant.[22] This shows that the maternal perception of the crying has an important impact on the problem.

Finally, mothers of excessively crying infants perceive the crying as worrisome, have congruent reactions, but remain rather strong psychologically, while these infants in fact cry relatively often. These mothers seem to be patient, caring, warm, responsive mothers, who have a high tolerance level. In a Dutch study, Van der Boom studied mothers of irritable infants and concluded that these mothers were indeed very sensitive and responsive.[23] Home observations have also shown parents of 'crying babies' to be highly responsive and sensitive, but differences between moderate and excessive criers were not described.[24,25]

Within the moderately crying group, the infants' age at enrolment, which is higher than that of the excessively crying infants, is related to the depression scale of the GHQ – 28, 'fatigue' on the POMS – R and all scales related to social dysfunction of the SSL-D. This relation is not found within the mothers of excessively crying infants.

It is quite conceivable that the longer the crying persists, the stronger it can influence maternal mood. Since the age of enrolment of moderately crying infants is higher than the age of the excessively crying infants, this can explain that the results measured by the mood scales do not differ between these two groups of mothers.
We feel that this might indicate that parents of excessively crying infants perceive their infant's crying behaviour as difficult, but it does not disturb their balance completely. Furthermore, psychological disturbances do not seem to be the cause of the amount of crying, but rather the effect, since the differences between mothers of excessive and moderate criers are relatively small. A similar conclusion is proposed by Papoušek and Von Hofacker.[10]

For practical reasons, we did not interview fathers and only asked mothers a few questions to assess by proxy the father's experience of pregnancy and their marital relation. Fathers of excessively crying infants only have a more negative experience of their wives pregnancy according to their partners. All other items are comparable in both groups.
A supportive marital relationship during pregnancy and the postpartum period is known to facilitate adjustment to the new parenting roles and to predict interactional competence with the infant.[26] Moreover, the quality of the marriage has been found to have impact on the parent-infant relationship.[27,28]
Psychological stress and partnership conflicts have been found to be associated with excessive crying of infants[10] and, in the subgroup with excessive crying according to Wessel's rule of three's, problems in family interaction persist longer.[29]
In a large Dutch cohort study, mothers who perceived their infants as excessive criers had a higher depression score compared to fathers, while in the group who cried excessively according to Wessel's criteria, the depression scores were about the same when comparing mothers to fathers.[4] Furthermore, paternal depression was correlated to excessive crying according to Wessel's criteria and, surprisingly, maternal depression was correlated to perceived excessive crying.

This, again, demonstrates that perception of excessive crying and mood are interrelated and influence the seriousness of the problem, as perceived by parents and presented by parents to health care workers.

Another finding of our study is that in both intervention groups (regularity vs. regularity combined with swaddling) the crying reduces equally in excessively crying infants and moderately crying infants.
Furthermore, we found evidence that infants who cry moderately and excessively share the same cry features and almost the same mothers, but the differences seem to be a matter of degree. The clinical phenomenon is not solely determined by infant characteristics but parental vulnerabilities are also part of the picture.[30]

Conclusion

As presented to clinicians, 'problem infant crying' is not just an infant condition, but parents' evaluations and vulnerabilities are involved as well.[31] The assessment of actual infants crying should be part of every study, preferably using behaviour cry diaries. Furthermore, the role of parental depression, social supports and other vulnerabilities of mothers and fathers should be assessed. More research is needed into the father-mother-child triage.
Mothers of excessive and moderate criers differ very little psychologically. Parents of excessively crying infants perceive the crying behaviour of their child as difficult, but it does not throw them out of balance completely. Furthermore, psychological disturbances do not seem to be the cause of the amount of crying but rather its effect, since the differences between mothers of excessive and moderate criers are relatively small.

The two interventions (regularity vs. regularity with swaddling added) do not differ in effect when measuring parental perception between excessively and moderately crying infants. In both groups the excessive crying reduces equally; whether this is due to the intervention, placebo effect or the natural history of crying, cannot be determined from this study. Other studies however have shown that regularity has a beneficial effect compared to care as usual. Nevertheless, this suggests that both interventions are applicable for the total range of crying infants whose parents seek help.

References

1. Crying in babies. In: Brugman E, Reijneveld SA, Hollander-Gijsman ME den, Burgmeijer RJF, Radder JJ. Gauging in the child health care 1997-1998 (41-45). Leiden: TNO Prevention and Health, 1999
2. Sleuwen BE van, L'Hoir MP, Engelberts AC, Busschers WB, Westers P, Blom MA, Schulpen TWJ, Kuis W. Comparison of behaviour modification with and without swaddling as interventions for excessive crying. J Pediatr 2006;149:512-7
3. Wessel, MA, Cobb JC, Jackson EB, Harris GS, Detwiler AC. Paroxysmal fussing in infancy, sometimes called "colic". Pediatrics 1954;94:322-32
4. Van den Berg MP, van der Ende J, Crijnen AAM Hofman A, Jaddoe VWV, Mackenbach JP, Moll HA, Tiemeier H, Hengeveld MW, Verhulst FC. Maternal and paternal depression during pregnancy and excessive infant crying. The Generation-R study. Thesis Erasmus University, 2006
5. Barr RG. Infant crying and colic: it's a family affair: invited commentary. Inf Ment Health 1995;16:218-20
6. Lester BM, Boukydis CFZ, Garcia-Coll CT, Holl WT, Peucker M. Infantile colic: cry characteristics, maternal perception of cry, and temperament. Infant Behav Dev 1992;15:15-26
7. Barr RG, Kramer MS, Boisjoy C, McVey-White L, Pless IB. Parental diary of infant cry and fuss behaviour. Arch Dis Child 1988;63:380-7
8. Lehtonen LA, Rautava PT. Infantile colic: natural history and treatment. Curr Probl Pediatr 1996;26:79-85
9. Räihä H, Lehtonen L, Huhtala V, Saleva K, Korvenranta H. Excessively crying infant in the family: mother-infant, father-infant and mother-father interaction. Child Care Health Dev 2002;28:419-29
10. Papoušek M, von Hofacker N. Persistent crying in early infancy: a non-trivial condition of risk for the developing mother-infant relationship. Child Care Health Dev 1998;24:395-424
11. Räihä H, Lehtonen L, Korvenranta H. Family context of infantile colic. Inf Ment Health 1995;16:206-17
12. Barr RG, Rotman A, Yaremko J, Leduc D, Francoeur TE. The crying of infants with colic: a controlled empirical description. Pediatrics 1992;90:14-21
13. Lucassen LB, Assendelft WJ, Gubbels LW, Eijk JT van, Douwes AC. Infantile colic: crying time reduction with a whey hydrolysate; a double blind, randomized placebo-controlled trial. Pediatrics 2000;106:1349-54
14. Rautava P, Helenius H, Lehtonen L. Psychosocial predisposing factors for infantile colic. BMJ 1993;307:600-4
15. Høgdall CK, Vestermark V, Birch M, Plenov G, Toftager-Larsen K. The significance of pregnancy, delivery and postpartum factors for the development of infantile colic. J Perin Med 1991;19:251-7
16. Huizink AC, Mulder EJH, Robles de Medina PG, Visser GHA, Buitelaar JK. Is pregnancy anxiety a distinctive syndrome? Early Hum Dev 2004;79:81-91

17. Van den Bergh B. The influence of maternal emotions during pregnancy on fetal and neonatal behavior. Pre-Peri-Nat Psych J 1990;5:119-30
18. Koeter MWJ, Ormel J. General Health Questionnaire [Dutch Translation]. Lisse: Swets & Zeitlinger BV, 1991
19. Ark, LA van der, Marburger, D, Mellenbergh GJ, Vorst HCM, Wald F. Revised Profile Of Mood States (POMS – r). Manual and Justification [In Dutch]. Nijmegen: Berkhout Nijmegen BV, 1995
20. Sonderen E van. Social Support List – Interactions (SSL – I) and Social Support List – Discrepancy (SSL – D). Manual [In Dutch]. University of Groningen, 1993
21. Central Bureau for Statistics (CBS). Birth, height and weight at birth and breastfeeding rates [In Dutch]. Voorburg, 2006
22. Pauli-Pott U, Becker K, Mertesacker T, Beckmann D. Infants with "colic"-mothers' perspectives on the crying problem. J Psychosomatic Research 2000;48:125-32
23. van den Boom DC. Neonatal irritability and the development of attachment: observation and intervention. Thesis, Leiden State University, 1988
24. Alvarez M. Caregiving and early infant crying in a Danish community. J Dev Behav Pediatr 2004;25:91-8
25. St James-Roberts I. Conroy S, Wilsher K. Links between maternal care and persistent infant crying in the early months. Child Care Health Dev 1998;24:353-76
26. Lester BM, Boukydis CFZ, Garcia-Coll CT, Peucker M, McGrath MM, Vohr BR, Brem F, Oh W. Developmental outcome as a function of the goodness of fit between the infant's cry characteristics and the mother's perception of her infant's cry. Pediatrics 1995;95:516-21
27. Belsky J, Russell AI. Marital and parent-child relationships in family of origin and marital change following the birth of a baby. A retrospective analysis. Child Dev 1985;56:342-49
28. Lamb ME, Elster AB. Adolescent mother-infant-father relationships. Dev Psychol 1985;21:768-73
29. Lehtonen L, Cormally S, Barr RG. 'Clinical pies' for etiology and outcome in infants presenting with early increased crying. In: Crying as a sign, a symptom and a signal. Eds: Barr RG, Hopkins B, Green J. MacKeith Press, USA, 2000
30. Murray L, Stanley C, Hooper R, King F, Fiori-Cowley A. The role of infant factors in postnatal depression and mother-infant interactions. Dev Med Child Neurol 1996;38:109-19
31. Papousek M, Wurmser H, von Hofacker N. Clinical perspectives on unexplained early infant crying: challenges and risks for mental health and parent-infant relationships. In: New evidence on unexplained early infant crying: its origins, nature & management (pp. 289-316). Eds: Barr RG, St James-Roberts I, Keefe MR. London: MacKeith Press/ Cambridge University Press, 2001

Appendix A

Groups of variables used to assess characteristics of parents (according to Rautava and Høgdall).[14,15]

MARITAL RELATIONSHIP

Are you pleased to be a woman? (1=very pleased, 5=not pleased)
Was your partner able to transpose himself into your state of mind during pregnancy? (1=very much, 5=not at all)
Did your partner understand your way of thinking during pregnancy? (1=very much, 5=not at all)
Has there been any change in your relationship during the pregnancy? (1=for the better, 5=for the worse)
Has there been any change in your sexual relationship during the pregnancy? (1=for the better, 5=for the worse)

WAY OF LIFE DURING AND ADAPTATION TO PREGNANCY

Did you feel to be on your own during the course of your pregnancy? (1=no, not at all, 5=yes, completely)
Did you experience emotional support during pregnancy? (1=yes, a lot, 5=no, none)
Did you feel socially isolated from other during the course of your pregnancy? (1=no, never, 5=yes, often)
Did you have less contact with friend than usual? (1=no, equally, 5=yes, much less)
Use of cultural service – How many times annually do you go to a cinema, theatre, concert, an athletic competition or match, a library, lectures, or public discussion? Then we calculated the yearly use for every respondent.

PHYSICAL HEALTH, EVENTS, AND SYMPTOMS IN PREGNANCY

Are you suffering from fatigue, digestive disturbances, heartburn, swelling, constipation, varicose veins, itching skin, tenderness or aches at the pubic symphysis, and severe contractions? (Asked after delivery, when the excessive crying occurred. The individual pregnancy trouble points were then calculated.)
Have you experienced migraine, headache, aching joints, pain in wrists, elbow joints and shoulder joints, back pain, swelling in legs or feet, and skin disorder. (Individual symptom points then calculated.)
Are you suffering from diabetes, epilepsy, hypothyroidism, hyperthyroidism, hypertension, allergic disorder, rheumatism, celiac disease, mental disorder, recurrent urinary tract infections, back disorders, other disorders? Please list.
Amount of sick leave during pregnancy was calculated as days.
What is your estimation about your own health? (1=healthy, 5=ill)

Have you felt that your work has been more tiring during the pregnancy than before? (yes/in some degree/no)

PARENTS' EXPERIENCE DURING CHILDBIRTH AND PROJECTIONS OF SELF IMAGE

How did you experience your pregnancy? (1=positive, 5=negative)
How did your partner experience your pregnancy? (1=positive, 5=negative)
Did you feel that your pregnancy was easier/more difficult than you had expected?
Did you feel that your pregnancy went according to your expectations or not at all?
Was your pregnancy a period of happiness/horror?

Mothers were asked about their experience of delivery evaluations and labour pains.
Mothers were also asked:
Did you feel that your delivery was easier/more difficult than you had expected?
Was your delivery a period of happiness/horror?
How did you experience your labour pains? (1=positively, 5=very negative)
How did your partner experience your labour pains? (1=positively, 5=very negative)
Did you feel that your childbed was easier/more difficult than you had expected?
Was your childbed a period of happiness/horror?
Did you feel insecure after during baby was born?

In the first weeks after your delivery, did you feel that you needed outside help? (1=no, not at all, 5=yes, a lot)
Were you able to let other people take care for your baby? (1=yes, very often, 5=no, never)
Do you feel limited in your freedom since your baby is born? (1=no, not at all, 5=yes, very much)

General discussion and conclusions

8

Bregje E. van Sleuwen

Introduction

Modern Western society is very complex. Thirty years ago parents were told to adhere to very strict childrearing schedules. This approach turned to "infant demand" care in the sixties and seventies. Nowadays, a child rearing approach with on-the-hour feeding schedules appears to be the norm, but with a lack of regularity and an enormous overload of stimuli. This approach might partly be the cause of the common problem of excessive crying of infants in many Western societies. Babies that cry excessively may also sleep less. Infants that persist in excessive crying (after four months of age) often have sleep problems at an older age.[1] Furthermore, excessive crying might lead to frustration and undesirable parental reactions. Consequently, there is a strong need for support for the wide range of excessively crying infants and their parents.
Crying is one of the first challenges parents are confronted with in the communication with their newborn infant. The normal pattern of crying has been well established and has age-dependent and diurnal features.[2] Crying usually begins to increase about two weeks after birth, peaks around 6 weeks, and declines to a more stable but lower levels by the fourth month.[3] When an infant fusses or cries excessively, communication can be disrupted and parents are challenged even more.

Theoretical model of excessive crying

A substantial amount of papers has been published on the pathogenesis and treatment of excessive crying. In earlier days it was referred to as "colic", which implies a medical cause. Even after several systematic reviews, neither uniform treatment nor "the cause" of excessive crying is identified.[4]
Keefe proposed a theoretical model that conceptualised infantile excessive crying from a developmental psychobiologic perspective. Infant state is conceptualised as a level of arousal continuum ranging from deep, quiet sleep to awake and aroused lusty crying. In the proposed model the excessively crying infant has a disorganized or immature sleep-wake cycle that underlies the excessive crying and difficulty in initiating sleep.[5] The process underlying the observed behavioural manifestations is seen as a dysfunction in the infant's ability to self-regulate state or sleep-wake cycles due to central nervous system immaturity rather than the gastrointestinal system disorder that the term colic seems to imply. This behaviour instability may be exacerbated by parental inconsistency and environmental disruptions. Keefe states: "From this parent-infant interaction framework, irritable infants are viewed as sensitive and more easily over-stimulated by busy, chaotic environments. As they become overwhelmed and fatigued, they cannot self-soothe or reduce their arousal level sufficiently to fall asleep. Parents, while very concerned, may actually be reinforcing the irritable behaviour pattern by inconsistent attempts at strategies that are not contingent with the infant's unclear signals and erratic cues".

Swaddling

Some form of infant restraint seems to be a worldwide childcare practice. Cross cultural and historical findings show that swaddling or bundling is used to attempt to reduce tension for parents with an infant that cries excessively; swaddled infants are perceived to be calm and sleep more. Swaddling might have an effect on the child, but it could also have an effect on parents. Chisholm wrote: "Although infants conceivably could learn that crying when swaddled (on the cradleboard) had no effect on the mother, it seems more likely that mothers have learned that the swaddling (cradleboard) itself will stop the child's crying".[6]
If the last holds true, other ways of infant restraint should be as effective in calming infants, as long as their situation is predictable. Firm tucking in of an infant might have the same calming and behaviour regulating effect.
Level one research into the efficacy of swaddling to reduce excessive crying was lacking so far, but substantial research supported its efficacy in achieving two other desired endpoints: cry reduction in infants with neonatal cerebral insults[7] and improvement of sleep[8]. The efficacy of swaddling is under-researched and study findings about infant crying are variable and contradictory, mostly because of different definitions for excessive crying that make comparison difficult.[9] However, in the Netherlands we noticed an increasing tendency to swaddle infants in order to curb their excessive crying. We felt it was undesirable to allow this trend to continue as there was no proven efficacy and there was no safe protocol for the use and the discontinuation of swaddling. To the best of our knowledge, no randomised trial has ever been performed into the added effect of swaddling on excessive crying. After exchanging swaddling experiences with people of several cultures, knowing the risk of prone sleeping for cot death, which is not self-evident when swaddled, and realizing the effect of the shaking baby syndrome that is often preceded by excessive crying, a study into the effect of swaddling was a challenge.

The randomised controlled trial

We chose to compare two approaches: a) a standardized approach consisting of regularity and predictability and, b) the same approach supplemented with swaddling. In the randomised trial, the intention was to include 400 infants. Each family was supported for 3 months by 3 visits to a health care worker and 3 telephone calls by the healthcare nurse. For organizational purposes they were also called by telephone three times by the researcher. Infants, who were included, were not older than 12 weeks, were healthy, and had no increased risk of hip dysplasia. The study group was stratified according to region, age and the amount of excessive crying.

A validated 24-hour diary and a maternal perception scale of their infant's crying were used. [10,11] Infant crying, both within normal range and excessive crying, follows a clear pattern after birth. Shortly after birth it increases in frequency and reaches a peak at six weeks of age, and then decreases in frequency, with or without outside help. Therefore, any study into cry reducing strategies must take into account the natural history of infant crying. Another factor that must be taken into account is the fact that the empathy and attention that is invariably associated with any intervention has great influence on the parental perception of crying and can strengthen the parents in their role as parents and thus influence the problem per se. In our study the follow up encompasses the period when the crying would significantly change or decline by itself. However, this was taken into account by the study design (RCT) and the analysis: the natural history of crying exerts its influence equally in both intervention groups making comparison valid. As both intervention groups received the same number of telephone and face to face contacts, the influence of the attention generated by the study is also comparable in both groups.
Including a control group would have theoretically enabled us to study the difference between an intervention and the natural history of crying. Parents in the Netherlands with excessively crying infants were supported in many different ways, both in type of strategy chosen and intensity of support by health care professionals. There was no care as usual for excessively crying infants. Many, perhaps even most parents did seek help, however. Any Dutch control group would therefore have been very heterogeneous and most probably not have reflected the "natural history" of crying. A control group would anyhow have contact with the research health nurse and would also have to fill out the 24-hour diaries, which is an intervention by itself, even if no extra attention or advice about the crying is given. This is affirmed by Keefe, where participating parents in the control group began to comment on the benefits derived from having the evaluation team members visit their home.[12] Not using diaries would make the data incomparable and our research invalid. Also, it did not seem appropriate not to offer any supportive strategy for parents seeking help when they perceive the problem as being significant. We know it to be associated with serious sequelae such as child abuse and shaken baby syndrome. Due to these considerations we therefore chose not to include a control group.

The approach chosen for our study as described in Appendix I was based on earlier (clinical) work. The baby care introduced to all parents in this study is comparable to the one described by Hofacker, Wolke, Keefe and in the Netherlands by Blom.[12-15] It consists of a recurrent pattern of baby care and not an on-the-hour or rigid time schedule. This exists of sleeping, waking up, directly followed by feeding, positive interaction/cuddling and playing with the parent, playing alone, preferably in a playpen, and, as soon as the baby shows signs of weariness (yawning, whining, rubbing its eyes, overactive behaviour) the baby is put into bed awake and tucked in tightly with sheet and blanket(s) that cover the child up to his chin, while feet to foot (feet against the footboard of the babies bed).

All parents received this advice of regularity and stimulus reduction and then they were randomised whether or not swaddling was added. The swaddling method as used in the randomised controlled trial is developed by Blom. In her book she extensively describes which swaddling cloths should be used, which fabric, the size of the two cloths and the clothes the child should wear underneath.[15] In chapter 5 this is summarized.

Aim of the randomised controlled trial

The aim of the study was to define if swaddling, when supplementary to regularity and stimulus reduction, has an added effect on the course of excessive crying. The second goal was to determine in what amount swaddling influences parental perception of their infant's cry.

Results of the randomised controlled trial

After the first intervention week, crying reduced with 42% in both groups, with 50% after 2 weeks, and with 75% after eight intervention weeks. Actual time spent crying decreased from an average of 148 minutes crying per day at baseline to 36 minutes crying per day at the end of the intervention period. The decrease of crying during the intervention for infants aged 4, 6, 8 and 10 weeks at baseline compared to the normal crying curve is visualised in Figure 1. After the first day of intervention, behaviour modification with swaddling decreased the mean duration of crying from baseline (mean decrease 30-40 minutes) while behaviour modification without swaddling increased it (mean increase of 20-25 minutes). After seven days the two groups did not differ. Parental perception of the worrisomeness of their infant's crying did not differ between both intervention groups. We concluded that adding swaddling to behaviour modification in infant care (i.e., regularity of care and stimulus reduction) did not reduce excessive crying in healthy infants less than 13 weeks of age at randomisation. Subgroup analyses on age groups however show that in infants 0 – 6 weeks of age at baseline, adding swaddling caused a significantly greater decrease in crying. However, this difference was only on average 10 minutes per day. In infants aged 7 – 12 weeks at baseline the effect was reversed: adding swaddling was actually contra-reproductive, as it caused about 10 minutes more crying per day.

Parental psychological factors

In 1957, Breslow stated the hypothesis that "emotional instability on the parts of the parent" might be causal in the development of excessive crying in infants, particularly for those in the group who did not respond to formula changes.[16] Another author, cited by Breslow, noted that excessive crying frequently improved "by substituting a calm nurse for an agitated mother".

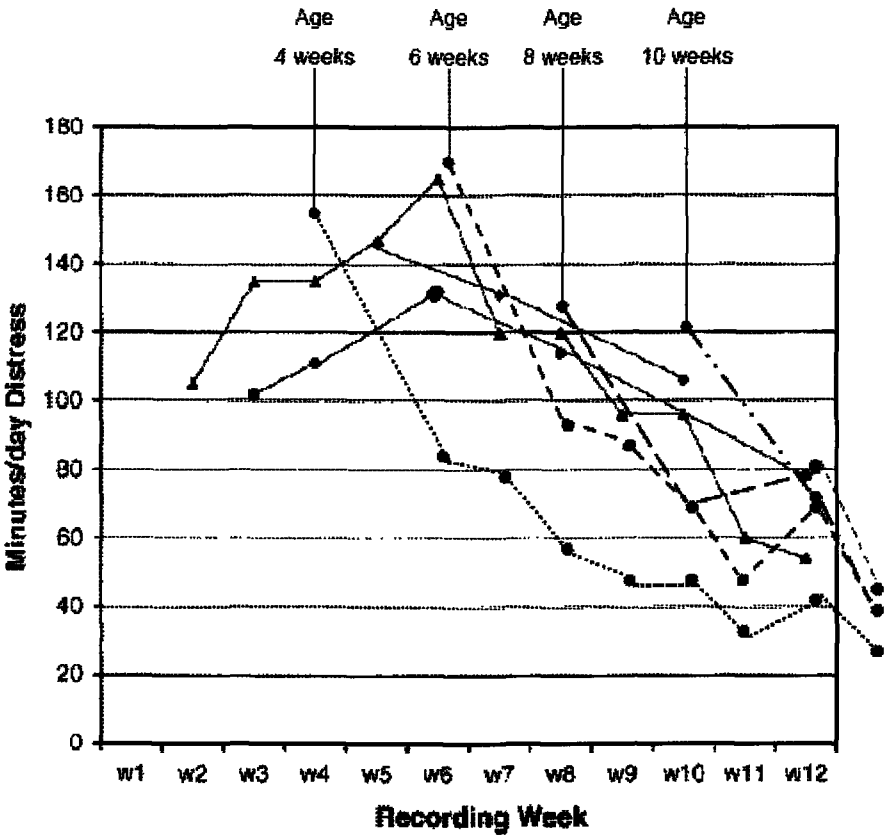


Figure 1. Crying amounts and patterns from three North American studies compared to the decrease of crying during the intervention for infants aged 4, 6, 8 and 10 weeks at baseline. Triangles: data from Brazelton (1962); circles: data from Hunziker and Barr (1986); diamonds: data from Kramer et al. (2001).

Reprinted from Barr RG, Trent RB, Cross J. Age-related incidence curve of hospitalized SBS cases: convergent evidence for crying as a trigger to shaking. Child Abuse Negl 2006;30:7-13.

"Of course, these comments reflect a time when it was thought that a number of unexplained conditions, such as autism, could somehow be caused by parental factors", commented Frohna in 2006.[4] Indeed, fifty years after Breslow's study, the cause and treatment of infantile excessive crying remains elusive.

The design of this study allows a cautious conclusion about prenatally predicting factors for excessive crying. Only a few differences are found between mothers of moderate (parental perception of excessive crying, but crying <3 hours a day according to a 24-hours baby day diary) and excessively crying infants (crying >3

hours/day according to a 24-hour baby day diary). Mothers of moderate crying infants showed more anxiety during pregnancy, which might explain why they asked for support by attending this study, while, objectively seen, their infants did not cry excessively. Mothers of excessively crying infants seem to react congruent to the problem they have to face, i.e. their infants cry a lot. The stress they experience and their psychological state seem to be the effect rather than the cause of the excessive crying.
The effect of both interventions did not differ for the group of infants that cried excessively compared to those that cried moderately. Also effects were similar both for mothers who score in the clinical range on psychological measurements as for mothers who do not. We conclude that our approach is applicable for a large group of moderately to excessively crying infants, independent of parental make-up (Chapter 7 and 8). Parents are positive about both interventions. If supportive care is needed the method of baby care can be offered. Swaddling gives no added benefit but can be used as an alternative strategy if parents so desire for example due to ethnic preferences. It should then be ensured that the swaddling method used is safe and discontinued before the infant starts turning, again due to safety issues. These strategies are appreciated and well tolerated so that using them is reasonable even if their efficacy has not been established compared to the natural history of crying.

Parental support

Brackbill observed that upset newborns often require a layering on of multiple soothing inputs (from arm restriction to holding to rocking) to reduce agitation.[17] Our current study shows that the simple regularity model helps parents to regain emotional power. This simple approach is easy to apply for parents when supported by their health nurse.
Parents of excessively crying infants desperately seek help. Over recent years, evidence has mounted that excessive infant crying can cause significant morbidity and mortality. It has been associated with nursing failure, marital discord, postpartum depression, Sudden Infant Death Syndrome, Shaken Baby Syndrome, excessive evaluation or treatment of gastroesophageal reflux disease, and disordered bonding.[18] During our approach we registered the amount of crying. A reduction of 42% was observed in the first week, but as stated previously we cannot compare this to the natural history of crying. Therefore, we cannot conclude if this is in fact due to the intervention itself. However, the intervention is much appreciated by parents and health nurses. Reduction of excessive crying may offer renewed opportunities for bonding, loving and attachment of parents and their infant.
It is important not to lose sight of the father, because when the misbalance is restored, all family members will profit.

Mothers of excessively crying infants are, except from the fact that they are exhausted, psychologically relatively healthy. They experience their infant's crying as devastating, but it does not throw them out of balance completely. However, most

mothers perceived the social support given in this study as insufficient. Social support has been described to mediate the negative effect of excessive crying in many mothers. In the Netherlands there is a well-organized system of maternity care, which supports newborn parents in the first week after the birth. This care is given at home and supervised by midwives. The maternity care workers are able to assess the social support by which the families are surrounded. This information should be added to the case history that is handed over to the well baby clinic after the postnatal period and used to target extra support.

Conclusion

The interventions costs little time, the support offered is much appreciated by the parents and the advice is easy to follow. One can conclude that a repetitive fixed pattern of baby care, which is not an on-the-hour schedule, in a contemporary busy Western world seems to support parents of excessively crying infants. Swaddling has no added benefit.
Our study, as concluded by Long, adds further doubt about swaddling effectiveness.[9]

Strengths and weaknesses

Parental assessment of their infant's amount of crying was used as inclusion criterion for this study. Because of the known fact that parents regularly overestimate the amount their infant cries[19], stratification was performed on crying less or more than 5 hours per day. Of all 348 infants whose diary recordings were complete and whose parents estimated their amount of crying as 5 hours or more per day at baseline (n=140, 40%), only 80 (57%) cried for more than 3 hours per day according to their 24-hour diary. When parental estimation of the amount of crying was split into less or more than 3 hours per day, 176 (67%) cried less than 3 hours per day, while their parents estimated their amount of crying as 3 hours or more.

In this study we did not include a control or 'routine care'-group. The reasons for this choice and the consequences that choice carries were discussed previously.

Implementation of the research findings and practical implications for clinical management

The difference of crying reduction in the first week between the two strategies in our study has practical implications for the implementation. In the regularity group the amount of crying increased on the first day of the intervention, and in the regularity group where swaddling was added, the crying decreased on the first day. From the fourth day on there was no significant difference in the amount of crying between both intervention groups.

Based on our results, a nationwide campaign was launched in 2005 through well baby clinics to effectuate a standardized support for parents who seek help with their excessively crying infants. Although there is no evidence from our study that enables us to compare its effect to that of the natural history of crying, the rapid reduction in crying we described in the first week and the positive and effective way the parents and health nurses perceived the intervention are the basis of our recommendation to use the program if help is asked for. More than 3500 health care workers received education and followed a workshop on both the approach of regularity and of a safe swaddling technique (see flowchart, Appendix II).
For infants whose crying is excessive and/or is perceived as problematic by their parents and/or physicians, health care nurses at well baby clinics propose regularity and reduction of stimuli as a first choice. As described in the randomised trial, crying should reduce within one week. When this approach fails, swaddling may be added to this approach. Sometimes, swaddling can be introduced to the regularity approach from start onwards, when parents are extremely stressed and/or tired and it is assumed by the healthcare nurse that they will not be able to deal with the short-term increase in crying that is seen when the regularity and stimuli reduction is applied. The Dutch National Institute for Public Health and the Environment (RIVM) has provisionally approved the new maternal and child nursing standard of regularity in excessively crying infants in the Netherlands.[20]

From research it is known that there are currently no clinically useful ways to predict which baby will cry a lot. St. James-Roberts outlined a standard approach of how to support parents with an excessively crying infant.[21] "Excessive crying is a parental complaint about infant crying, so parents as well as infants are always involved. Accurate assessment of infant behaviour is a vital first step. The use of a 24-hour diary or other instruments to measure the amount, the pattern and the nature of crying will generate diagnostic information and may be helpful in providing parents with reassurance and insight. Parent's responses to crying will vary and some will find it particularly difficult to cope with. The collection of information to identify maternal depression, social support, and other sources of vulnerability should be a core part of the primary workup. A physician should always rule out medical causes. After this the focus should be on providing parents with information, advice and support, tuned to local circumstances. The following issues should be addressed when counselling parents."
Important elements, as described by St. James-Roberts, could be supplemented with elements of the Dutch approach:

- "Examining the notion that crying means that there is something 'wrong' with a baby of this age and introducing alternatives – such as the possibility that it signals a reactive or vigorous baby[21];
- Viewing the first three months of infancy as a developmental transition, which all babies go through more or less smoothly[21];

- Reassuring parents that it is normal to find crying aversive and discussing the dangers of abuse and 'shaken baby syndrome'[21];
- Discussing ways of containing and minimising the crying, and highlighting positive features of the baby[21];
- Considering the availability of supports and the development of coping strategies which allow individual parents to take time out and to 'recharge their batteries'[21];
- Empowering parents and reframing the first three months as a challenge, which they can overcome, with positive consequences for themselves and their relationships with their babies[21]."
- Informing parents about the natural crying curve, the research into regularity, predictability and the small extra effect of swaddling for the young age group (2-7 weeks).
- Handing out the booklet about regularity and predictability and (if necessary) swaddling.
- Telling them if applicable that swaddling should be withdrawn before the age of six months, or earlier when a child attempts to turn to the prone position. A swaddled child in prone position has a high risk of cot death.
- Offering support if swaddling is necessary and continues this until the child is unswaddled permanently.
- Continuing to monitor infant and parents, preferably until the problem is over.[21]

Challenges for the future, and possible new avenues for research

A link with obesity?

Sufficient evidence exists that lack of sleep, or a short sleep duration, is a risk factor for overweight and childhood obesity.[22-24] Short sleep duration at the age of 30 months predicts obesity in seven-year old infants. Taheri postulates that loss of sleep at this young age may alter the hypothalamic regulation of appetite and energy expenditure. We pose that enlarging knowledge of parents about the self-regulating capacities of their child and supporting parents' self-efficacy in handling crying and sleep problems, and offering child rearing support, might be factors in the fight against obesity. In the Netherlands the approach of regularity and stimuli reduction is being implemented in all well-baby clinics and is appealing to most parents.
This might be a (first) step in the prevention of overweight and childhood obesity, but further exploration is needed.

Follow-up study

A follow-up study is being executed to evaluate long-term effects of both interventions used in our study at an average infant's age of 3–5 years. In this

follow-up study, parents are asked to fill out a questionnaire on several items, such as the Child Behavioural Checklist (CBCL 1,5 – 5 years), the Nijmegen Parenting Stress Index – short version (NOSI – K), some scales from the Parenting Dimensions, the general infant development, and weight and length of the participating infants. At this moment, we have reached more than 70% of all participants and preliminary analysis indicate that, at the age of 3 – 5 years, trial-infants scored significantly more positive on the CBCL – item 'sleep problems' when compared to the CBCL norm-sample.[25] This is a promising finding, because many parents of former excessively crying infants report sleeping problems during toddlerhood.[26-33]

Primary prevention

Employing regularity and uniformity in daily infant care might also a measure to prevent unrest, excessive crying and sleeping problems[34-39]. Clinical experience shows that the regularity approach might prevent unrest and excessive crying when started about 1 – 2 weeks after birth.[15] To determine if the regularity approach is suitable for prevention of unrest, sleeping problems, and excessive crying, further research is needed.

Relation between breastfeeding and swaddling

In the Netherlands exclusive breastfeeding until the age of six months is promoted and breastfeeding rates are rising but still low. The latest breastfeeding rates show that 79% of the mothers initiate breastfeeding, but at the age of 3 months around 35% and at the age of six months only about 25% of the mothers exclusively breastfeed their infant.[40] Restless infants are not able to drink efficiently and a mother might assume that she lacks sufficient milk, which is an often-used argument when breastfeeding fails. Many parents in this study reported giving extra (formula)feeds to facilitate their infant's sleep, 61% of (partly) formula-fed infants were offered several different types of formula before entering the trial and 12% had even been introduced to a third or fourth type of formula. Further exploration into the relation between regularity (which can be explained as on-demand feeding and not as an on-the-hour schedule), with or without swaddling added, and breastfeeding is needed.

Persistent excessive infant crying

Complementary to the approach for excessively crying infants as described in our study, many Dutch child healthcare centres offer video-feedback intervention in addition to the intervention of regularity (sometimes supplemented with swaddling).[41] This supports parents and increases the interaction between parents and children.
In addition to this, for depressed mothers and their babies an effective early mother-baby intervention method that includes video-feedback is available in Dutch

community mental health centres. In a randomised controlled trial, a group of depressed mothers of 1 – 12 month old infants received either an intervention programme (8 – 10 home visits) or parenting support by phone (three telephone contacts with a child therapist). Compared to the control group, maternal sensitivity, child responsiveness towards the mother, and child's involvement with the mother in interactions, significantly improved in the intervention group. Follow-up assessment at six months after the end of the intervention shows that the difference in the quality of the mother-child interaction between the intervention and control groups was retained. The infants in the experimental group also had significantly higher scores for attachment security and socio-emotional competence than infants in the control groups.[42]

Barr[26] and Papoušek[27] earlier described a 'persistent mother-infant syndrome', where in a subgroup of infants and parents, early excessive crying may not resolve but evolve to having problems in multiple behavioural domains, such as crying that persists beyond 4 months of age, sleeping, and feeding problems. Especially for this group of parents and their infants, additional support that includes video-interaction training might be a valuable intervention. Research into the added effect of video-interaction training in a group of long-term excessively crying infants will shortly start.

References

1. Wolke D, Rizzo P, Woods S. Persistent infant crying and hyperactivity problems in middle childhood. Pediatrics 2002;109:1054-60
2. Lehtonen L, Gormally S, Barr RG. 'Clinical pies' for etiology and outcome in infants presenting with early increased crying. In: Barr RG, Hopkins B, Green JA (Eds.). Crying as a sign, a symptom, and a signal. MacKeith Press, London 2000
3. Räihä H, Lehtonen L, Huhtala V, Saleva K, Korvenranta H. Excessively crying infant in the family: mother-infant, father-infant and mother-father interaction. Child Care Health Dev 2002;28:419-29
4. Frohna JG. 50 Years ago in The Journal of Pediatrics. J Pediatr 2007;10.1016/j.jpeds.2006.09.006
5. Keefe MR. Irritable infant syndrome: theoretical perspectives and practice implications. ANS Adv Nurs Sci 1988;10:70-8
6. Chisholm JS. Swaddling, cradleboards and the development of children. Early Hum Dev 1978;2/3:255-75
7. Ohgi S, Akiyama T, Arisawa K, Shigermori K. Randomised controlled trial of swaddling versus massage in the management of excessive crying in infants with cerebral injuries. Arch Dis Child 2004;89:212-216
8. Gerard CM, Harris KA, Thach BT. Physiologic studies on swaddling: An ancient child care practice, which may promote the supine position for infant sleep. J Pediatr 2002;141:398-403

9. Long T. Adding swaddling to behaviour modification in infant care did not reduce excessive crying in healthy infants <13 weeks of age at randomisation. Evid Based Nurs 2007;10:42
10. Barr RG, Kramer MS, Boisjoly C, McVey-White L, Pless IB. Parental diary of infant cry and fuss behaviour. Arch Dis Child 1988;63:380-7
11. Lester BM, Boukydis CFZ, Garcia-Coll CT, Holl WT, Peucker M. Infantile colic: cry characteristics, maternal perception of cry, and temperament. Infant Behav Dev 1992;15:15-26
12. Keefe MR, Barbosa GA, Froese-Fretz A, Kotzer AM, Lobo M. An intervention program for families with irritable infants. MCN 2005;30:230-6
13. Hofacker N von, Papoušek M, Jacubeit T, Malinowski M. Rätsel der Säuglingskoliken. Ergebnisse, Erfahrungen und therapeutische Interventionen aus der "Müncher Sprechstunde für Schreibabies". Monatsschr Kinderheilkd 1999;147:244-53
14. Wolke D, Gray P, Meyer R. Excessive infant crying: a controlled study of mothers helping mothers. Pediatrics 1994;94:322-32
15. Blom MA. Crying and Restlessness in Babies. A Parent's Guide to Natural Sleeping. Edinburgh: Floris Books, 2005
16. Breslow L. A clinical approach to infantile colic: a review of ninety cases. J Pediatr 1957;50:196-206
17. Brackbill Y. Cumulative effects of continuous stimulation on arousal levels in infants. Child Dev 1971;42:17-26
18. Karp H. Swaddling and excessive crying. J Pediatr 2007;151:e2-e3
19. van der Wal MF, van den Boom DC, Pauw-Plomp H, de Jonge GA. Mothers' reports of infant crying and soothing in a multicultural population. Arch Dis Child 1998;79:312-7
20. ActiZ. Interventions of excessive crying in infants [In Dutch]. Bunnik, 2007
21. St James-Roberts I. Managing the crying of 1-3 month old infants in western cultures. Presentation conference Paediatric Section of the Royal Society of Medicine, 2002
22. Boere-Boonekamp MM, L'Hoir MP, Beltman M, Bruil J, Dijkstra N, Engelberts AC. Overweight and obesity in preschool children (0-4 years): behaviour and views of parents [In Dutch]. Ned Tijdschr Geneeskd 2008;152:324-30
23. Taheri S. The link between short sleep duration and obesity: we should recommend more sleep to prevent obesity. Arch Dis Child 2006;91:881-4
24. Sleuwen BE van, Engelberts AC, Boere-Boonekamp MM, L'Hoir MP. Behaviour modification and swaddling as interventions to improve sleep: A link with obesity? Arch Dis Child jan 3rd 2007 (electronic pages)
25. Bruin FE de. Excessively crying infants: Follow-up 5 years after intervention. Utrecht University, 2007
26. Barr RG. Crying in the first year of life: good news in the midst of distress. Child Care Health Dev 1998;24:425-39

27. Papoušek M, von Hofacker N. Persistent crying in early infancy: a non-trivial condition of risk for the developing mother-infant relationship. Child Care Health Dev 1998;24:395-424
28. Kries R von, Kalies H, Papoušek M. Excessive crying beyond 3 months may herald other features of multiple regulatory problems. Arch Pediatr Adolesc Med 2006;160:508-11
29. Wake M, Morton-Allen E, Poulakis Z, Hiscock H, Gallagher S, Oberklaid F. Prevalence, stability, and outcomes of cry-fuss and sleep problems in the first 2 years of life: prospective community-based study. Pediatrics 2006;117:836-42
30. Rao MR, Brenner RA, Schisterman EF, Vik T, Mills JL. Long term cognitive development in children with prolonged crying. Arch Dis Child 2004;89:989-92
31. Wolke D, Rizzo P, Woods S. Persistent infant crying and hyperactivity problems in middle childhood. Pediatrics 2002;109:1054-60
32. Rautava P, Lehtonen L, Helenius H, Silanpaa M. Infantile colic and family three years later. Pediatrics 1995;95:43-7
33. Wade S, Kilgour T. Extracts from "Clinical Evidence": Infantile colic. *BMJ* 2001;323:437-40
34. Symon BG, Marley JE, Martin AJ, Norman ER. Effect of a consultation teaching behaviour modification on sleep performance in infants: a randomised controlled trial. Med J Aust. 2005;182:215-8
35. St. James-Roberts I, Sleep J, morris S, Owen C, Gillham P. Use of a behavioural programme in the first 3 months to prevent infant crying and sleeping problems. J Paediatr Child Health 2001;37:289-97
36. Kerr SM, Jowett SA, Smith LN. Preventing sleep problems in infants: a randomized controlled trial. J Adv Nurs 1996;24:938-42
37. Pinilla T, birch LL. Help me make it through the night: behavioral entrainment of breast-fed infant's sleep patterns. Pediatrics 1993:91:436-44
38. Adair R, Zuckerman B, Bauchner H, Philipp B, Levenson S. Reducing night waking in infancy: a primary care intervention. Pediatrics 1992;89:585-8
39. Wolfson A, Lacks P, Futterman A. Effects of parent training on infant sleeping patterns, parent's stress and perceived parental competence. J Consult Clin Pscyhol 1992;60:41-8
40. Lanting CI, van Wouwe JP. Peiling melkvoeding van zuigelingen 2005: Borstvoeding in Nederland en relatie met certificering door stichting Zorg voor Borstvoeding. Rapport KvL/JPB/2006.017. Leiden: TNO, 2006
41. Eliëns M. Images of babies. Video-home training and video-interaction in vulnerable babies [in Dutch]. Uitgeverij SWP, Amsterdam 2005
42. Doesum K van. An early preventive intervention for depressed mothers and their infants, its efficacy and predictors of maternal sensitivity. Thesis, Radboud University Nijmegen, 2007

Summary

9

Bregje E. van Sleuwen

In Chapter 1, a brief introduction is presented on normal infant crying, its definition, prevalence and the possible causes, followed by the predictors, consequences and long-term effects of excessive crying. A review is given on interventions that have attempted to reduce excessive crying. Furthermore, both historical and current beliefs on swaddling are discussed, as well as the build-up to the randomised trial. Finally, the aim of the study and the outline of the thesis are described.

In Chapter 2, the possible beneficial and detrimental effects of swaddling are described in a systematic review, based on best evidence in current literature. A historical review and the effect of swaddling on sleep and arousal, temperature control, motor development, Sudden Infant Death Syndrome (SIDS), respiratory infections, rickets, Developmental Dysplasia of the Hip (DDH), pain control, crying behaviour, breastfeeding and postnatal weight and swaddling start and duration are described.

Beneficial effects of swaddling are:

- Less startles, less arousal, but a higher arousability and longer sleep in healthy infants. When swaddled, infants seem to return to sleep on their own more easily.
- An improved neuromuscular development, less physiological distress, better motor organization and more self-regulatory ability during weighing in preterm infants.
- Decrease in excessive crying in infants with cerebral damage when compared to massage.
- Swaddling during heelstick makes preterm infants return to their baseline heart rate and oxygen saturation measures more quickly and soothes preterm infants after pain. Swaddling is less immediately effective than pacifier use, but is also less subject to rebound compared to a pacifier.
- No influence was found on breastfeeding parameters such as number and duration of breastfeeds, amount of ingested milk and total duration of breastfeeding time.

Possible detrimental effects of swaddling are:

- An increased risk for the development of developmental dysplasia of the hip (DDH), especially when the child is swaddled in extension and adduction. This detrimental effect of swaddling is related to the misapplied use of this practice. Swaddling in a manner that hips and knees can freely move might not have this risk increasing side effect, but more studies are needed to test this hypothesis.
- A risk for overheating. As soon as traditional swaddling practices are introduced in a Western society with (centrally) heated houses and highly

insulating bedding, one should be aware of the possible side effect of these practices and inform parents about the risks this practice might imply.

- An increased risk for sudden infant death syndrome (SIDS), when swaddled in prone position or when the infant is able to turn to the prone position when swaddled. The risk for SIDS seems to be increased by sleeping in a prone position when swaddled and by swaddling with the head covered. Swaddling in a supine position seems to be protective, although this diverges with the swaddling-method used. Up to a certain age, swaddling might hinder turning prone.
- An added risk of vitamin D deficiency, due to lack of sunlight. There is no indication that the vitamin D deficiency is such that it can lead to rickets.
- A risk for acute respiratory infections. Four studies in non-Western countries describe a higher risk for respiratory infections that seem to be related to the tightness of swaddling.
- A delayed weight-loss recovery in swaddled infants who are shortly separated from their mothers after birth or placed in a nursery, and who receive supplements.

Different techniques of swaddling have been used in many cultures. When not used properly, swaddling can be a dangerous intervention. It can be used safely, however, if accompanied by advice about sleeping position, the manner the child is swaddled (head free), the amount of (extra) bedding and the tightness of the swaddle around chest, hips and knees. Preterm infants can benefit from swaddling as well, although the technique is different from postnatal swaddling of full-grown infants.

In Chapter 3, differences in infant care practices related to cot death in Turkish, Moroccan and Dutch families in the Netherlands are described. The use of swaddling in the study group as well as the estimated prevalence rates of mothers being swaddled during their own childhood is presented. In the Netherlands 80% of the Turkish and Moroccan parents were swaddled in infancy. Nowadays fifteen percent of the Turkish children and 20% of the Moroccan children living in the Netherlands are swaddled, while around 6% of the Dutch infants are. Observation and clinical practice shows that these ethnic groups often swaddle inappropriately; the legs and hips are swaddled very tightly in extension and adduction. It is important to support and educate these parents about safe swaddling.

In Chapter 4, the main study is described. In the Netherlands, excessive crying occurs in 5-15% of all infants, depending on what definition is used. A variety of different interventions can be launched to reduce the crying, but in the Netherlands there is no fixed strategy. Sometimes parents may employ potentially dangerous interventions to reduce the crying, for example placing the infants in a prone sleeping position or co-sleeping with their infant. Swaddling might be an effective method to reduce excessive crying but it has never been systematically investigated.
A standardized approach, which consists of offering regularity and stimulus reduction, was compared with an experimental group, which received the same approach, supplemented with swaddling during all sleeping periods. The design was a randomised controlled trial, with a follow-up of 12 weeks. Babies all over the Netherlands were referred to a specially trained health nurse at 4 locations: Utrecht, The Hague, Arnhem, and Raalte/Zwolle. Included were 398 healthy infants who were ≤12 weeks and 6 days of age, had a gestational age ≥32 weeks, and cried excessively (>3 hours per day for ≥3 days/week) according to parents, doctors, or healthcare workers.
After a one-week baseline period during which all infants received hypoallergenic formula (or in case of breastfeeding, a maternal diet free of milk, egg, wheat, or nut products), infants were excluded if crying decreased to <2 hours/day from day 3/4 onwards, or if parents perceived the crying not excessive anymore. Infants at risk of DDH because of familial predisposition or breech position in the last month before birth were also excluded.
The intervention consisted of behaviour modification in infant care with (n=194) or without (n=204) swaddling. The behaviour modification focused on regularity of care and stimulus reduction. Parents provided a recurrent pattern of infant care based on a sequence of sleeping, feeding, positive parent-infant interaction, and then the infant was placed awake and alone in a playpen. Upon the first signs of tiredness, the infant was put to bed, while still awake and tucked in tightly with sheets and blankets, and a new cycle began. Swaddling involved wrapping infants in 2 rectangular cloths (shoulders and arms wrapped tightly; less tightly from the hip down to allow for leg flexion and abduction) during all sleeping periods. Parents completed 24-hour diaries for a consecutive period of 7 days, and then 1 fixed day a week. Nurses made 15 minute supportive calls on days 2 and 5 and a 30 minute call at week 9; the project manager made a 30 minute supervision call at weeks 5 and 12. The outcome included parents' perceptions of crying (Cry Perception Scale) and amount of crying (24-hour diary).

The patient follow-up was 96% (mean age 7.9 weeks). After the first intervention week, crying reduced with 42% in both groups, with 50% after 2 weeks, and with 75% after eight intervention weeks. Crying decreased from an average of 148 minutes crying per day at baseline to 36 minutes crying per day at the end of the intervention period. After the first day of intervention, behaviour modification with swaddling decreased the mean duration of crying from baseline (mean decrease 30-40 minutes) while behaviour modification without swaddling increased it (mean increase of 20-25 minutes). After seven days the two groups did not differ. We concluded that adding swaddling to behaviour modification in infant care (i.e., regularity of care and stimulus reduction) did not reduce excessive crying in healthy infants less than 13 weeks of age at randomisation. Subgroup analyses on age groups however show different results. In infants 2 – 7 weeks of age at baseline, adding swaddling caused a significantly greater decrease in crying. However, this difference was only on average 10 minutes per day. In infants aged 8 – 13 weeks at baseline the effect was reversed: adding swaddling was actually contra-reproductive, as it caused about 10 minutes more crying per day.
The behavioural intervention costs little time, the support offered is very much appreciated by the parents and very easy to follow. We conclude that a repetitive fixed pattern of baby care supports parents of excessively crying infants in reducing the excessive crying of their infant within several days. In general, swaddling has no relevant added benefit.

In Chapter 5, the amount of infants that were diagnosed cow's milk allergic by a provocation test is investigated by means of a follow up study. Furthermore, the determinants that might discriminate between favourable and non-favourable reactions to hypoallergenic diet are identified.
Forty-seven infants (10.6%) of all 398 infants that cried excessively reacted positively to a hypoallergenic diet given during the first week of the study and before any intervention was started. After two and a half years, 35 infants with a mean age of 35 months were located for follow up. Their parents reported more maternal asthma, paternal hay fever, and parental atopy. Furthermore, their parents smoked more often than parents of infants who did not react to the hypoallergenic diet, they less often exclusively breastfed, and they were less stressed during first trimester of pregnancy.

Strongly advised by the research group to consult the doctor at the well-baby clinic, only 34% of the parents of excessively crying infants with a suspected cow's milk allergy did return to the well-baby clinic for a re-challenge, re-elimination test. Of these 6 infants (50%) were diagnosed to have a cow's milk allergy. Parents of infants who react positively to a hypoallergenic diet need extra support to perform a re-challenge, re-elimination test. Apparently they are so relieved that the child's crying has reduced, that they avoid a possible relapse into the recurrent pattern of crying and accompanying stress.

In Chapter 6, maternal psychological and social state are compared between mothers of excessively crying infants, mothers of infants who react positively to one-week hypoallergenic diet, and control mothers of non-excessively crying infants. Furthermore, the relation between high and low maternal scores (clinical versus non-clinical range) on several psychological measures and the outcome of both interventions were investigated.
Mothers of excessively crying infants perceived more negative feelings and less support during pregnancy than mothers of infants who reacted positively to a hypoallergenic diet.
Compared to controls, mothers of excessively crying infants, contrary to mothers of infants who react positively to one-week hypoallergenic diet, reported having more physical complaints during pregnancy and more symptoms of illness during pregnancy.
When looking at psychological scores, the fact that there are so few differences between mothers of infants who react positively to a week hypoallergenic diet and mothers of excessively crying infants, suggest that the higher proportion of clinical scores in both groups compared to controls is due to the excessive crying rather than a cause of the crying. The efficacy of both interventions, regularity and regularity with swaddling added, was not associated with psychological differences of the mothers.

In Chapter 7, psychological characteristics were investigated in mothers of excessively crying infants, moderately crying infants and controls. Excessive crying is crying for more than three hours per day according to a 24-hour diary, moderate crying is defined as excessive crying according to parents but not according to a 24-hour diary.
The efficacy of two interventions to reduce excessive crying (regularity compared to regularity with swaddling added) was compared for excessively crying infants and moderately crying infants.
Mothers of excessively crying infants did not differ in pregnancy related anxieties compared to mothers of moderately crying infants or controls, although mothers of moderately crying infants were more anxious than control mothers. On the Cry Perception Scale, mothers of excessively crying infants experience the crying as more worrisome than mothers of moderately crying infants and control mothers. Mothers of moderately crying infants perceived crying significantly more often as 'spoiled' compared to control mothers and to mothers of excessively crying infants.
Mothers of excessively crying infants score more negative on the POMS – R: 'depression' scale than mothers of moderately crying infants. Both interventions (regularity vs. regularity with swaddling added) are as effective for moderately as well as excessively crying infants.

The cry behaviour of excessively crying infants is perceived as difficult by their parents, but it does not imbalance them completely. Furthermore, psychological disturbances do not seem to be the cause, but rather the effect of the amount of crying, as only the item 'depression' shows a higher score and not pregnancy related anxiety. As the two interventions (regularity vs. regularity with swaddling added) do not differ in effectiveness between mothers of excessively and moderately crying infants, this suggests that both interventions are applicable for the total range of mothers whose baby cries, regardless of the amount of crying.

In Chapter 8, a general discussion and conclusion on the efficacy of swaddling on excessive crying is presented. In a concluding section, practical implications and directions for future research are described.

In Appendix I, practical guidelines for both interventions, as used in the randomised trial (Chapter 4), are set out for health nurses. The consultation process, the frequency of support, and the content of both interventions are fully described. Examples of infants who participated in the study are given and it is demonstrated how the interventions accustom the infants to a stable sleep-wake rhythm, in fact how they learn a new, appropriate behaviour. The effect is that the infants develop their self-regulating ability and are at ease again. They cry less, sleep sufficiently, drink efficiently and in adequate amounts, they are no longer overtired and are able to fall asleep on their own again. The infants are also more able to play on their own and to enjoy cuddling. Through these interventions a vicious circle of asking and paying attention in the wrong way, for example by entertaining or consoling a child that is tired and needs sleep, is broken. The endurance of the parents may be restored, which may further promote a healthy attachment.
The technique of swaddling that meets all safety requirements is presented. In view of the increasing ability to roll over on its tummy, it is necessary to have stopped swaddling before the infant is six months old or at any moment when a child makes an attempt to turn to the prone position. Advice on how to cease the swaddling is fully described.

The main findings are:

- The regularity approach is based on two principles: offering the infant predictability and developing its self-regulating ability. The approach will reduce excessive crying and helps bring about a stable sleep-wake rhythm.
- A likely cause of excessive crying is overtiredness. By restoring/establishing a stable sleep-wake rhythm, overtiredness disappears, crying decreases and sleep increases.
- Nurses can easily teach regularity and uniformity in everyday childcare in a series of concrete advices to parents.

The following conclusions are based on the research and literature review as presented in this thesis:

1. The use of swaddling seems to have some beneficial effects, but one should be reserved to present it as a universal childcare habit as it also has possible detrimental effects.
2. Swaddling in the Netherlands is more widespread among people with non-western cultural habits, although the use of swaddling is becoming more common as intervention for excessive crying.
3. A repetitive fixed pattern of baby care, with or without swaddling, supports parents of excessively crying infants and reduces excessive crying within several days.
4. Infants aged 2 – 7 weeks at baseline show a greater decrease in crying during the eleven weeks of the intervention when they are swaddled, infants aged 8 – 13 weeks at baseline show a greater decrease in crying when regularity without swaddling is applied. The actual difference, although statistically significant is in fact very small, ten minutes per day, and therefore the clinical relevance is dubious.
5. Only 34% of the parents of excessively crying infants who react positively to one-week hypoallergenic diet returned to the well-baby clinic for a re-challenge, re-elimination test.
6. Parents of infants who react positively to one-week hypoallergenic diet report more parental atopy, they smoked more often than parents of infants who did not react to the hypoallergenic diet, and they less often exclusively breastfeed.
7. Mothers of excessively crying infants perceived more negative feelings and less support during pregnancy than mothers of infants who reacted positively to one-week hypoallergenic diet.
8. Mothers of excessively crying infants reported having more physical complaints and more symptoms of illness during pregnancy than control mothers.
9. Mothers of moderately crying infants were more anxious in pregnancy and they perceived their infant's crying as more 'spoiled' than control mothers and mothers of excessively crying infants.
10. Mothers of excessively crying infants perceived the crying of their infant as more worrisome than mothers of moderately crying infants or controls and they score more negative on the 'depression'-scale of the POMS – R than mothers of moderate criers.
11. Both interventions (regularity and regularity with swaddling added) are as effective for moderately as well as excessively crying infants. Furthermore, their effectiveness is not dependant on psychological characteristics of the mothers.

Nederlandse Samenvatting

9

Bregje E. van Sleuwen

Normaal huilen

Het huilen van een baby is één van de eerste en meest indrukwekkende vormen van communicatie tussen ouder en kind. Tijdens de eerste levensmaanden is een voorspelbaar patroon van huilen waarneembaar. Dit wordt ook wel de normale huilcurve genoemd. Vanaf de geboorte neemt het huilen geleidelijk toe totdat het een piek bereikt op de leeftijd van 6 – 8 weken (gemiddeld 2 – 2,5 uur per dag). Daarna neemt het huilen weer af tot het rond de leeftijd van 3 maanden een niveau heeft bereikt wat stabiel blijft de rest van het eerste levensjaar (1 – 1,5 uur per dag). Daarnaast huilen de meeste kinderen vaak meer in de avonduren. Deze normale huilcurve lijkt onafhankelijk te zijn van de manier van opvoeden, in tegenstelling tot de hoeveelheid huilen later in het eerste levensjaar.

Excessief huilen

De meest gehanteerde definitie van excessief huilen is opgesteld door Wessel en wordt wel de "regel van drie" genoemd: een gezond en goed gevoed kind die perioden van jengelen of ontroostbaar huilen heeft die minimaal 3 uur per dag duren en ten minste gedurende 3 dagen per week voorkomen. Excessief huilen kan gezien worden als een objectief te meten fenomeen, maar ook als een perceptie van ouders, waarbij de ouders bepalen of zij het huilen als abnormaal ervaren. Om de hoeveelheid huilen, jengelen, slapen en periodes van tevreden spelen te meten, bestaat er een gevalideerd "24-uurs dagboek". De waarneming van ouders over het huilen van hun kind is te interpreteren met een "huilperceptieschaal".
In Nederland huilt ongeveer 2 – 2,5% van de zuigelingen excessief wanneer de criteria van Wessel gehanteerd worden. Wanneer ouders wordt gevraagd of hun baby in de afgelopen week op tenminste één dag meer dan 3 uur gehuild heeft, antwoordt 8.6 – 13% bevestigend.

Doel van dit onderzoek

Inbakeren wordt in toenemende mate toegepast in Nederland, zonder dat bekend was of dit van invloed was op het huilen van baby's en op de perceptie van ouders over het huilen van hun kind. Het doel van dit onderzoek was tweeledig:

- Vaststellen of inbakeren, wanneer dit wordt toegevoegd aan een interventie waarbij regelmaat en prikkelreductie in de zorg van de baby centraal staan, een toegevoegde waarde heeft en het huilen reduceert.
- Vaststellen in welke mate het toevoegen van inbakeren de perceptie van ouders beïnvloedt.

Resultaten

In Hoofdstuk 1 wordt een korte introductie gegeven over normaal huilen, waarna de definitie, de prevalentie en mogelijke oorzaken, eventuele voorspellers, consequenties en lange termijn gevolgen van excessief huilen worden beschreven. In een literatuuroverzicht worden interventies en het mogelijke effect hiervan op de reductie van huilen weergegeven. Daarnaast worden historische en huidige inzichten over inbakeren besproken, evenals de aanloop naar dit onderzoek. Als laatste wordt het doel van het onderzoek uiteengezet en wordt een overzicht gegeven van de inhoud van dit proefschrift.

In Hoofdstuk 2 wordt op basis van een systematisch literatuuroverzicht een overzicht van de mogelijke positieve en negatieve effecten van inbakeren, gebaseerd op evidentie uit de literatuur, gegeven. Het is een historisch overzicht en het effect van inbakeren op de slaap, het ontwaken, de temperatuurcontrole, de motorische ontwikkeling, wiegendood, luchtweginfecties, de ontwikkeling van rachitis (Engels Ziekte), dysplastische heupontwikkeling, pijn, huilgedrag, borstvoeding en postnataal gewicht wordt beschreven. Bovendien wordt de methode van het inbakeren behandeld.

Mogelijke positieve gevolgen van inbakeren zijn:

- Gezonde ingebakerde kinderen hebben minder motorische onrust en worden minder vaak wakker tijdens de slaap, maar zijn daarentegen wel gemakkelijker te wekken. Ze lijken langere perioden achter elkaar te slapen, maar kinderen die ingebakerd zijn, vallen gemakkelijk weer zelf in slaap wanneer ze wakker zijn geworden.
- Prematuur geboren kinderen maken een snellere neuromusculaire ontwikkeling door, hebben minder fysiologische stress, een betere organisatie van de motoriek en een beter ontwikkeld zelfregulerend mechanisme tijdens het wegen wanneer ze ingebakerd zijn.
- Een afname van huilen bij cerebraal beschadigde kinderen wanneer inbakeren vergeleken wordt met massage.
- Inbakeren tijdens een hielprik zorgt ervoor dat prematuur geboren kinderen eerder terugkomen op hun uitgangsniveau qua hartritme en O_2-niveau en het troost prematuur geboren kinderen na pijn. Inbakeren geeft hierbij een minder directe reactie in vergelijking met het gebruik van een fopspeen, maar zorgt ook voor een minder snelle terugval.
- Er is geen invloed gevonden op borstvoeding parameters, zoals het aantal voedingen en de duur van de borstvoeding, hoeveelheid ingenomen melk en totale duur van de borstvoedingsperiode.

Mogelijke negatieve gevolgen van inbakeren zijn:

- Een verhoogd risico op een dysplastische heupontwikkeling, met name wanneer het kind in extensie en adductie ingebakerd wordt. Dit negatieve gevolg van inbakeren is groter wanneer de methode niet correct wordt toegepast. Inbakeren op een manier dat de knieën en heupen vrijelijk kunnen bewegen, lijkt geen verhoogd risico te geven op de ontwikkeling van de heup, maar meer onderzoek op dit gebied is noodzakelijk.
- Een verhoogd risico op oververhitting. Sinds inbakeren is geïntroduceerd in de Westerse landen waar (centrale) verwarming en warmte-isolerend beddengoed gebruikt wordt, moet men alert zijn op het mogelijke bijeffect van dit gebruik en dienen ouders geïnformeerd te worden over dit risico.
- Wanneer baby's ingebakerd op de buik worden gelegd of ingebakerd naar de buik rollen, verhoogt dit het risico op wiegendood. Een baby inbakeren met het hoofd bedekt, verhoogt het risico eveneens. Inbakeren in rugligging lijkt een beschermende werking te hebben, maar dit is afhankelijk van de methodiek van inbakeren die gebruikt wordt. Tot op een bepaalde leeftijd kan inbakeren het draaien naar de buik belemmeren.
- Een verhoogd risico op vitamine D-deficiëntie door een tekort aan zonlicht. Er zijn geen indicaties dat de vitamine D-deficiëntie zodanig is dat deze kan leiden tot rachitis.
- Een verhoogd risico op acute luchtweginfecties. Vier studies in niet-Westerse landen beschrijven een verhoogd risico op luchtweginfecties die gerelateerd lijken te zijn aan de stevigheid van inbakeren.
- Een uitgesteld herstel in gewicht na de geboorte wanneer kinderen vlak na geboorte in het ziekenhuis ingebakerd worden en een korte periode worden gescheiden van hun moeder of in de babyslaapzaal te slapen gelegd worden, en wanneer baby's worden bijgevoed.

In verschillende culturen worden diverse methoden van inbakeren gebruikt. Wanneer inbakeren niet op een juiste manier wordt toegepast, kan dit risicovol zijn. Het kan veilig toegepast worden wanneer het vergezeld gaat van advies over slaappositie, de techniek waarmee het kind wordt ingebakerd (hoofd dient vrij te blijven), de hoeveelheid beddengoed en hoe strak de doeken rondom de borst, knieën en heupen worden aangebracht. Prematuur geboren kinderen kunnen mogelijk profiteren van inbakeren, hoewel de techniek van inbakeren in deze groep verschilt van die bij à term geboren zuigelingen.

In Hoofdstuk 3 worden de verschillen in verzorgingsgewoonten tussen gezinnen met een Turkse, Marokkaanse en Nederlandse achtergrond in relatie tot het risico van wiegendood beschreven. De prevalentie van inbakeren in de studiegroep, evenals de geschatte prevalentie van het gebruik van inbakeren bij de ouders toen zij een baby waren, wordt gepresenteerd. Gedurende hun kindertijd werd 80% van de Turkse en Marokkaanse ouders zelf ingebakerd. Tegenwoordig wordt in Nederland 15% van de

zuigelingen met een Turkse achtergrond, 20% van de zuigelingen met een Marokkaanse achtergrond en ongeveer 6% van de Nederlandse zuigelingen ingebakerd. Observaties en klinische ervaring laat zien dat deze etnische groepen vaak een onveilige manier van inbakeren gebruiken; de benen en heupen worden strak en in extensie en adductie ingebakerd. Het is van enorm groot belang dat deze ouders goed geïnformeerd en begeleid worden bij het gebruik van een veilige manier van inbakeren.

Hoofdstuk 4 beschrijft de resultaten van de hoofdstudie. In Nederland komt excessief huilen bij 5 – 15% van de zuigelingen voor, afhankelijk van de definitie die wordt gebruikt. Er wordt een groot scala aan interventies aangeboden die huilen mogelijk kunnen reduceren, maar een eenduidige aanpak wordt momenteel niet toegepast. Ouders kunnen soms risicovol gedrag vertonen om het huilen te doen reduceren, zoals bijvoorbeeld de zuigeling op de buik te slapen leggen of hun baby bij zich in bed nemen om te slapen. Inbakeren als aanvulling op het bieden van regelmaat en voorspelbaarheid kan mogelijk een effectieve methodiek zijn om huilen te reduceren, maar het effect hiervan was nog niet systematisch onderzocht. Een gestandaardiseerde aanpak, bestaande uit het bieden van regelmaat, voorspelbaarheid en prikkelreductie wordt in deze studie vergeleken met een experimentele aanpak waarbij ouders dezelfde adviezen krijgen, aangevuld met het inbakeren van de zuigeling tijdens elke slaapperiode. Het onderzoek betrof een gerandomiseerd gecontroleerde trial (RCT) met een follow-up periode van 12 weken. Kinderen uit heel Nederland werden verwezen naar speciaal voor het onderzoek getrainde JGZ verpleegkundigen die gelokaliseerd waren in 4 regio's: Uitrecht, Den Haag, Arnhem en Raalte/Zwolle. In totaal werden 398 excessief huilende zuigelingen geïncludeerd (leeftijd ≤12 weken, minimale zwangerschapsduur 32 weken). In de eerste week, de baseline week, ontvingen alle zuigelingen een hypoallergeen dieet (Frisolac Allergycare of een aangepast dieet voor moeder in geval de moeder borstvoeding gaf, vrij van melkproducten, ei, noten, pitten en zaden). Wanneer na deze baselineweek het huilen gereduceerd was tot < 2 uur per dag en de ouders het huilen niet meer als excessief percipieerden, werd het kind geëxcludeerd van verdere deelname. Kinderen met een familiaire predispositie voor een dysplastische heupontwikkeling en kinderen die gedurende de laatste maand van de zwangerschap in stuitligging gelegen hadden werden uitgesloten van deelname. De interventie bestaande uit adviezen van regelmaat, voorspelbaarheid en prikkelreductie (n = 204) werd vergeleken met dezelfde interventie aangevuld met inbakeren (n = 194). Ouders boden de zuigelingen een herhalend verzorgingspatroon aan, gebaseerd op een cyclus van slapen, ontwaken, voeden, positieve interactie tussen ouder en kind, waarna de zuigeling in de box gelegd wordt (bij baby's > 4 – 6 weken) om even alleen te spelen. Bij de eerste vermoeidheidssignalen wordt het kind in bed gelegd en stevig toegedekt onder een laken en deken(s) en kreeg het de kans om zelf in slaap te vallen om de ontwikkeling van de zelfregulatie te stimuleren. Na ontwaken begon weer een nieuwe cyclus. Bij het inbakeren werd gebruik gemaakt van twee doeken, waarbij schouders en armen stevig ingebakerd werden. Vanaf de heupen naar de

tenen worden de doeken minder stevig aangelegd om flexie en abductie van de benen te kunnen bewerkstelligen. Ouders hebben gedurende een periode van 2 aansluitende weken een 24-uurs dagboek ingevuld. Na deze eerste twee weken werd het dagboek wekelijks op een vaste dag ingevuld. De verpleegkundigen hadden in de eerste interventieweek telefonisch contact met de ouders op dag 2 en 5 (15 minuten) en in week 9 (30 minuten). De projectcoördinator nam in week 5 en week 12 telefonisch contact op met de ouders (30 minuten). Deze twee gesprekken waren in principe geen steunende maar vooral organisatorische contactmomenten. De uitkomst van de interventie werd gemeten met een huilperceptieschaal en met het 24-uurs dagboek, waarmee onder andere de hoeveelheid huilen, jengelen en slaap werd gemeten.
In totaal heeft 96% van de ouders de volledige begeleiding van 3 maanden afgerond. De zuigelingen waren gemiddeld 8 weken oud bij inclusie (SD 2.52, min 2.6, max 13). Na de eerste interventieweek was het huilen in beide interventiegroepen gemiddeld met 42% afgenomen, na 2 weken met 50% en na 8 interventieweken met 75%. De huilduur bij aanvang van de baseline week van 148 minuten per dag nam af tot gemiddeld 36 minuten huilen aan het einde van de totale interventieperiode. Wanneer de adviezen van regelmaat aangevuld werden met inbakeren, is op de eerste interventiedag een daling van huilen van gemiddeld 30 – 40 minuten waarneembaar, terwijl bij gebruik van de adviezen van regelmaat zónder inbakeren het huilen op de eerste dag toenam met gemiddeld 20 – 25 minuten. Aan het einde van de eerste interventieweek was er geen significant verschil meer waarneembaar tussen beide interventiegroepen. Ouderlijke perceptie van het huilen van hun zuigeling was niet verschillend tussen beide interventiegroepen. We mogen hieruit concluderen dat het toevoegen van inbakeren op adviezen van regelmaat niet zorgt voor een grotere reductie in huilen bij gezonde zuigelingen tot de leeftijd van 13 weken. Subgroepanalyses bij verschillende leeftijdsgroepen laten echter een genuanceerder resultaat zien. Wanneer zuigelingen met een leeftijd van 2 – 7 weken bij inclusie adviezen van regelmaat èn inbakeren ontvangen, daalt het huilen significant meer dan in de groep baby's die ouder zijn dan 7 weken (gemiddeld 10 minuten minder huilen per dag). Zuigelingen in de leeftijdscategorie 8 – 13 weken bij inclusie laten juist het tegenovergestelde effect zien: zij huilen gemiddeld 10 minuten minder per dag wanneer zij alleen adviezen van regelmaat ontvangen, zonder inbakeren.
De adviezen van regelmaat, voorspelbaarheid en prikkelreductie kosten weinig tijd, de ondersteuning zoals geboden aan ouders, wordt erg gewaardeerd en is goed op te volgen. We concluderen dat een herhaald patroon van regelmaat en voorspelbaarheid ouders van excessief huilende zuigelingen ondersteunt en het huilen binnen enkele dagen reduceert. Het inbakeren heeft geen toegvoegde waarde; er is een klein effect meetbaar bij de jonge baby's, maar een verschil van 10 minuten per dag is klinisch niet relevant.

In Hoofdstuk 5 wordt de follow up beschreven van de zuigelingen die positief reageerden op de hypoallergene voeding in de baseline week.

Zevenenveertig kinderen (10.6%) van de 398 zuigelingen die excessief huilden bij inclusie reageerden positief op het hypoallergene dieet in de baseline week. Na 2,5 jaar zijn 35 kinderen met een gemiddelde leeftijd van 35 maanden geïncludeerd in een vervolgstudie. Hun ouders rapporteerden vaker astma bij moeder, hooikoorts bij vader en atopie bij één of beide ouders. Verder rookten ouders van kinderen die positief reageerden op de hypoallergene voeding vaker in vergelijking met ouders van excessief huilende zuigelingen die niet positief reageerden op de hypoallergene voeding, kregen deze kinderen minder vaak borstvoeding en hadden de moeders minder aan de zwangerschap gerelateerde angsten tijdens het eerste trimester van de zwangerschap. Tevens waren beide ouders jonger en waren de moeders minder vaak universitair geschoold. Bij het overgrote deel van de kinderen waarbij geen koemelkeitwitallergie is gediagnosticeerd middels eliminatie-provocatie-re-eliminatie, is het huilen wel afgenomen. Aan de ouders en kinderen die na de baseline meting werden geëxcludeerd, is nadrukkelijk gevraagd terug te gaan naar het bureau in verband met deze eliminatie-provocatie-re-eliminatie test. Slechts 34% heeft dit advies opgevolgd. Dit is enerzijds te begrijpen vanuit de ervaring van de ouders; de baby huilt niet meer en ze moeten er mogelijk niet aan denken dat zij opnieuw door zo'n stressvolle episode moeten. Anderzjjds betekent dit dat ouders meer steun nodig hebben om de test te doorlopen met hun kind. Zolang ouders dit niet doen, blijft het onduidelijk hoe groot het percentage kinderen is dat daadwerkelijk een koemelkeiwit allergie heeft.

In Hoofdstuk 6 wordt een vergelijking gemaakt tussen moeders van excessief huilende zuigelingen (n=382), moeders van zuigelingen die positief reageerden op *een hypoallergeen dieet (n=47) en controlemoeders van niet-excessief huilende* gezonde zuigelingen (n=369) op het gebied van psychologische en sociale factoren. Daarnaast is bij de moeders de relatie tussen hoge en lage scores (klinisch versus niet-klinisch) op verschillende psychologische schalen en het resultaat van beide interventies vergeleken.
Moeders van excessief huilende zuigelingen hebben tijdens de zwangerschap meer negatieve gevoelens en minder steun ervaren in vergelijking met moeders van zuigelingen die positief reageerden op een hypoallergeen dieet. In vergelijking met controlemoeders rapporteren moeders van excessief huilende zuigelingen, in tegenstelling tot moeders van zuigelingen die positief reageerden op een hypoallergeen dieet, vaker lichamelijke klachten en meer symptomen van ziekte tijdens de zwangerschap.
Het feit dat er zo weinig psychologische verschillen waarneembaar zijn tussen moeders van excessief huilende zuigelingen en moeders van zuigelingen die positief reageerden op een hypoallergeen dieet suggereert dat het hoge aandeel van beide groepen moeders in het klinisch gebied in vergelijking met controlemoeders een gevolg is van het excessieve huilen en niet de oorzaak van het overmatige huilgedrag. Het effect van beide interventies, regelmaat en regelmaat met inbakeren, bleek niet te zijn geassocieerd met psychologische verschillen tussen de drie groepen moeders.

In hoofdstuk 7 worden de psychologische verschillen tussen moeders van excessief huilende zuigelingen (n=102, 29%), moeders van matig huilende zuigelingen (n=246, 71%) en controlemoeders (n=369) beschreven. Excessief huilen wordt hierbij aangemerkt als huilen > 3 uur per dag volgens een 24-uurs dagboek. Matig huilen wordt gedefinieerd als huilen wat volgens ouders excessief is, maar volgens het dagboek < 3 uur.
Het effect van beide interventies (regelmaat en regelmaat met inbakeren) is vergeleken tussen de groepen kinderen die excessief en matig huilden.
Moeders van excessief huilende zuigelingen verschilden in vergelijking met moeders van matige huilers en controles niet wat betreft aan zwangerschap gerelateerde angsten, hoewel moeders van matig huilende zuigelingen angstiger waren dan controlemoeders. Moeders van excessief huilende zuigelingen percipiëren het huilen als zorgelijker (huilperceptieschaal) in vergelijking met moeders van matige huilers en controlemoeders. Moeders van matige huilers ervaren het huilen significant vaker als 'verwend' in vergelijking met controlemoeders en moeders van excessieve huilers.
Moeders van excessief huilende zuigelingen hebben een meer negatieve score op de POMS – R 'depressie' schaal in vergelijking met moeders van matige huilers. Beide interventies sorteren hetzelfde effect bij zowel matige als excessief huilende zuigelingen.
Het huilgedrag van excessief huilende zuigelingen wordt als moeilijk ervaren door hun ouders, maar ze raken er niet volledig door uit balans. Daarnaast lijken psychologische verstoringen niet direct de oorzaak maar eerder het gevolg te zijn van het overmatige huilgedrag. Alleen op het item 'depressie' wordt vaker negatief gescoord en niet op aan zwagerschap gerelateerde angsten. Het feit dat beide interventies geen verschil laten zien in effect tussen moeders van excessief en matig huilende zuigelingen, suggereert dat beide interventies toepasbaar zijn voor alle moeders wiens zuigeling overmatig huilt, onafhankelijk van de hoeveelheid huilen.

In Hoofdstuk 8 volgt de discussie en conclusie over het effect van inbakeren op de perceptie en de duur van excessief huilen. In een laatste paragraaf worden praktische adviezen en suggesties voor verder onderzoek gegeven.

In Appendix I worden de adviezen zoals genoemd in Hoofdstuk 4 uitgebreid beschreven.

Conclusies

De interventie kost weinig tijd, de begeleiding wordt zeer gewaardeerd door de ouders en de adviezen zijn eenvoudig op te volgen. We kunnen concluderen dat een patroon van regelmaat en voorspelbaarheid in de verzorging van een jonge baby, wat geen schema op tijd en klok is, in deze huidige drukke westerse maatschappij ouders van overmatig huilende zuigelingen lijkt te ondersteunen. Inbakeren heeft hier geen toegevoegde waarde.

Het verschil tussen beide interventiegroepen in de afname van huilen in de eerste week heeft gevolgen voor de adviezen voor de praktijk. In de regelmaatgroep neemt huilen toe op de eerste dag na start van de interventie, bij gebruik van regelmaat aangevuld met inbakeren neemt huilen af op de eerste interventiedag. Na 4 dagen was er echter geen verschil meer in de hoeveelheid huilen tussen beide interventiegroepen.
Gebaseerd op deze resultaten is een landelijke richtlijn opgesteld voor de begeleiding van excesssief huilende zuigelingen (concept-richtlijn 'Aanpak van excessief huilen bij zuigelingen', ActiZ 2007) en is in 2005 een landelijke scholing gestart voor jeugdgezondheidszorg-medewerkers om hen te informeren over de in dit onderzoek gebruikte aanpak voor ouders van baby's die excessief huilen. Hoewel in deze studie het effect van regelmaat, prikkelreductie en voorspelbaarheid niet kan worden vergeleken met het natuurlijk verloop van huilen omdat er geen controle groep ("care as ususal") is gebruikt, vormen de adviezen van regelmaat, voorspelbaarheid en prikkelreductie de basis van de aanbevelingen wanneer ouders om hulp vragen. De snelle reductie van huilen in de eerste week en de positieve ervaringen van ouders en hulpverleners vormen hiervoor de basis.
Bij kinderen van wie het huilen als overmatig wordt ervaren door ouders en/of hulpverleners wordt in eerste instantie regelmaat, voorspelbaarheid en prikkelreductie voorgesteld. Zoals beschreven in de studie, zou het huilen hierdoor binnen één week moeten afnemen. Wanneer deze aanpak niet of onvoldoende effect sorteert, is het mogelijk onder bepaalde voorwaarden inbakeren toe te voegen aan de aanpak van regelmaat. In een enkel geval kan inbakeren direct vanaf het begin toegevoegd worden aan de adviezen van regelmaat, maar slechts in die situaties waarin ouders extreem gespannen of oververmoeid zijn en de jeugdgezondheidszorgmedewerker inschat dat de ouders niet (meer) in staat zijn de toename in huilen op de eerste dag op te vangen. De landelijke richtlijn 'Aanpak van excessief huilen bij zuigelingen' (ActiZ, 2007) ligt momenteel ter vaststelling bij het RIVM. Inmiddels zijn deze adviezen in combinatie met een praktijkworkshops over de toepassing van de adviezen van regelmaat, voorspelbaarheid en prikkelreductie en de techniek en (contra-)indicaties van inbakeren overgedragen aan ruim 3.500 medewerkers van de jeugdgezondheidszorg.

Appendix I

Regularity and swaddling: Interventions that are applied when infants cry excessively

Maria A. Blom
Bregje E. van Sleuwen
Han de Vries
Adèle C. Engelberts
Monique P. L'Hoir

Submitted for publication

Abstract

The purpose of this article is to describe two healthcare nurse interventions that were developed to support parents whose infant cries excessively while also increasing the amount the infant sleeps. This article describes how the interventions can be used by nurses; the full study was published previously. Intervention 1 consists of advice to caregivers to bring about regularity and uniformity in their daily care of the infant and reduce external stimuli. Intervention 2 is the same advice but is accompanied by swaddling during all sleeping periods. The aim of both interventions is to habituate the infants to a stable sleep-wake rhythm, stop unacceptable crying behavior, and learn new, more appropriate behavior. Nurses could teach parents these interventions that are based on offering predictability, which allows infants to improve their self-regulating ability. We strive to let babies fall asleep on their own, sleep as much as they need to sleep, become well rested again, cry less, drink adequately, be able to play on their own and enjoy cuddling.

Callouts

- The regularity approach is based on two principles: the infant should be offered predictability, and the infant should be helped to develop his/her self-regulating ability.
- The behavioural regularity approach has been extensively used to support parents; during the program a rapid decrease in crying is seen in the first week and it helps to bring about a stable sleep-wake rhythm.
- A likely influencing factor of excessive crying is overtiredness. By restoring/establishing a stable sleep-wake rhythm, nurses can help teach parents how to prevent overtiredness, decrease crying and increase sleep.

Keywords

excessive crying, infantile colic, regularity, swaddling

Introduction

The purpose of this article is to describe interventions to help parents with excessively crying infants in order to help maternal and child nurses work with new parents more effectively. Excessive crying or colic occurs in 5% to 25% of all infants for some period in the first months of life (Roberts, Ostapchuk, & O'Brien, 2004). Several interventions have been studied that have been aimed at supporting parents with an excessively crying infant, with varied results (Roberts et al., 2004; Lucassen et al., 1998; Wade & Kilgour, 2001; Keefe, Barbosa, Frose-Fretz, Kotzer, & Lobo, 2005). Recently, van Sleuwen et al. (2006) published the results of a randomized trial to compare two interventions for excessive crying: (a) behavioral advice to caregivers to bring about regularity and uniformity in daily infant care and (b) the behavioral approach combined with swaddling the infant during all sleeping periods (van Sleuwen et al., 2007). This article describes the interventions in full; readers are also encouraged to access the complete report of the randomized trial (van Sleuwen et al., 2006) for detailed data analysis.

From Experience-Based to Evidence-Based Practice

Nurses are taught that practicing based on evidence is essential. Evidence (research) often results first from observations from clinical nurses and is then translated into clinical research in order to discover if the clinical observations hold up in controlled situations. This is what happened with our work with infants who cry excessively. The first author of this article is a maternal and child healthcare nurse at a Dutch healthcare center. She has closely observed babies with crying and sleeping problems and over the years has developed interventions for supporting families when infants cry excessively (Blom, 2001, 2005a, 2005b). By sharing insights with parents about the beneficial effects of regularity and uniformity in daily childcare on establishing or restoring a stable sleep-wake rhythm, many parents were able to reduce excessive crying in their infants. A somewhat similar approach emphasizing regularity was proposed by Hofacker, von Papoušek, Jacubeit, and Malinowski (1999) and investigated by Keefe et al. (2005).

Blom's positive experiences with regularity and swaddling, together with the approach by Hofacker et al., formed the basis of a randomized controlled trial in the Netherlands (van Sleuwen et al., 2006). In that 12-week study of 398 healthy infants aged 2 to 13 weeks who were reported to cry excessively, two interventions were compared: (1) regularity and uniformity in daily childcare and the simultaneous reduction of stimuli and (2) the same approach with swaddling added during all sleeping periods. In that sample of infants, the mean duration of crying initially assessed by the parents was 5.8 hours in each 24-hour period.

In a study by Keefe et al. (2005), the mean cry duration as assessed by two groups of American parents with children with a mean age of 5.1 and 10.4 weeks was similar (both groups: 5.9 hours per 24 hours). Results of the study by van Sleuwen et al. (2006) were presented by describing crying duration during the first week of interventions and then by subsequent weeks. Both of the interventions led to large differences in the average course of cry duration across the days of the first intervention week. For Intervention 1 (regularity in daily care and stimuli reduction), the amount of crying on the first day *increased* by about 20 to 25 minutes/24 hours on average but then *decreased* on the following 6 days by about 11 minutes/24 hours (Figure 1). In contrast, for Intervention 2 (regularity combined with swaddling), on the first day the amount of crying *decreased* by about 30 to 40 minutes, on the second day crying *increased* by about 10 minutes, and then, on the following 5 days it decreased by about 6 minutes per day (Figure 1). After 7 days, the differences in crying were not detectable by which intervention was being used. After the first week, both interventions resulted in an average reduction in crying of 42%, after 2 weeks there was a reduction of 50%, and after 8 weeks there was a reduction of 75%. When comparing the two interventions in the weeks *after* the first intervention week, swaddling seemed to decrease crying more in the younger infants (2-7 weeks old at the start of the intervention) than the older infants (8-13 weeks old at the start). The group differences within each age category were approximately 10 minutes. As there was no control group it is not possible to compare the efficacy of the interventions with "a wait and see" approach. However, it is possible to describe the predicted pattern of crying to the parents after starting intervention. This approach is much appreciated by parents and empowers them.

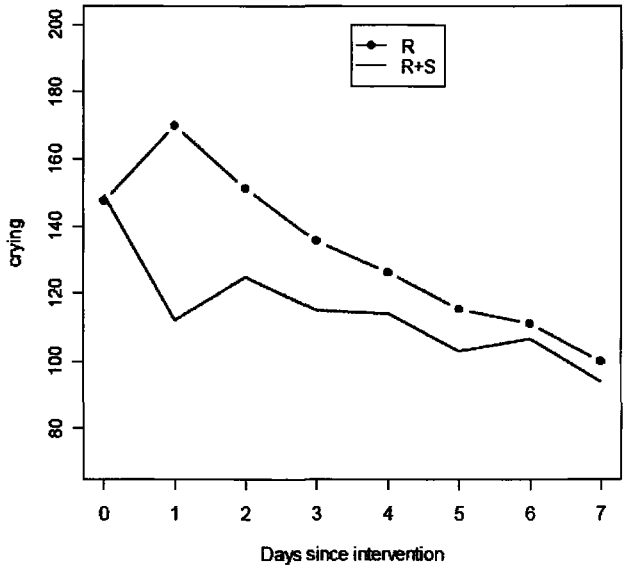


Figure 1. Average amount of crying (min/24 hours) during the first intervention week. R = regularity, R+S = regularity and swaddling

Interventions when infants are crying excessively

Our interventions are based on the concepts of *regularity and uniformity,* which means that these babies require a consistent, recurring pattern of sleeping, feeding right after waking, positive interaction/ cuddling and playing with a parent, playing on their own (preferably in a playpen), and being put to bed awake as soon as signs of weariness appear (e.g., yawning, whining, rubbing of eyes, overactive behavior). This approach offers the child predictability and the possibility to improve his or her self-regulating ability.

The behavioral components of the intervention include regularity in the *sequence* of the events surrounding sleep and feeding and uniformity in the *place* where the child sleeps and plays on his or her own. It should be emphasized that this does not mean a fixed time schedule but rather a consistent approach. The elements of the behavioral approach to excessive crying are shown in Box 1.

Helping Infants to Fall Asleep

As soon as signs of weariness are visible, the infant should be put to bed while still awake and tucked in tightly with sheet and blanket(s) that cover the child up to his or her chin, with his or her feet against the footboard of the crib. The ability to fall asleep on his or her own is a precondition for breaking through a pattern of dependence. Infants who cry excessively are often not used to falling asleep on their own. These infants need a real opportunity to learn to fall asleep and may need to cry themselves to sleep for 15 to 30 minutes. Parents can peek in on a child during this time but should take care not to be seen. Our experience has been that within 30 minutes the infant often falls asleep, although he or she was crying loudly just before.

Parents can be encouraged to use an egg timer to keep a realistic sense of time during this crying period, because it can seem like hours when an infant is crying. At the next bedtime, the baby often cries less. Some infants persist in crying for more than 30 minutes; then some consolation (e.g. soothing words) can be given without taking the child out of bed. If an infant remains unsoothable, parents should again start the cycle described previously (i.e., take the infant out of bed, feed him/her, and so on). When this approach is used, we have found that a child soon recognizes that being picked up is followed by being fed, and being put into bed is followed by going to sleep. Within this regular pattern the infant is fed shortly after waking up, because a well-rested infant drinks adequately, and a well-fed infant is able to play on his or her own contentedly without entertainment until he or she gets tired.

REGULARITY AND UNIFORMITY IN EVERYDAY CHILDCARE FOR INFANTS WHO CRY EXCESSIVELY

It is important that parents fully understand the whys and aims of the key elements in this approach, which will motivate them to put the new approach into practice.

Regularity in the sequence of events offers the infant predictability

- When the child wakes up he/she is fed right away, because a well-rested infant drinks adequately.
- After feeding, it is natural to cuddle and play with the infant, after which the infant has to play on his/her own in a fixed place.
- Upon the first signs of weariness, the infant should be put to bed while still awake. Intervals between daytime feeds are preferably 2 to 4 hours.

Uniformity in the place where events happen enhances predictability

- Playing on his/her own in a fixed place should be encouraged, preferably in the playpen.
- Infants should sleep in the same crib all the time (not in the playpen).

Reduction or avoidance of stimuli

- Silencing the radio, television, mechanically driven toys, and baby gym (if < 3 months old).
- Bouncy chair should be used only during feeding times, and many visits or outings should be discouraged.

Further advice to help the child to fall asleep on his/her own

- Offer a substantial boundary by tucking the baby in, up to the chin, while on his/her back.
- During the transition phase to a new sleep/wake rhythm, parents need to accept that an infant may cry for about 15 to 30 minutes before falling asleep.

When the intervention also includes swaddling

- Swaddle during every sleeping period and continue for at least 6 weeks.
- Stop swaddling after about 8 weeks, the time that the new rhythm is established.
- Stop swaddling immediately when the infant makes any attempt to turn prone.
- Stop swaddling at the latest when the infant becomes 6 months old.
- When swaddling is stopped, it is still appropriate to use a well-fitting "sleeping sack" that leaves arms free.

Note. From Blom, 2005b.

Box 1. The elements of the behavioral approach to excessive crying.

Reduction of Stimuli

It is important to reduce the stimuli around a newborn who cries excessively, which means avoiding stimulation from radio, television, mechanically driven toys, and baby gyms (if the child is < 3 months old), avoiding using a bouncy chair (except for feeding times), and avoiding continuous entertainment and many visits or outings.

Swaddling

Based on our research, swaddling adds no extra benefit to the behavioral intervention. However, some families are so distraught that the initial increase in crying that accompanies the behavioral approach can be too much for them, or sometimes there is a strong preference for swaddling. Swaddling can also be tried in selected cases if the behavioral approach is not successful. We then suggest that just before the infant is put into bed to sleep he or she should be swaddled while dressed in one layer of cotton clothing (allowing the child to be warm evenly over his/her body).

Swaddling is accomplished by wrapping him or her in two cloths from shoulders to toes in such a way that flailing arms and moving legs are limited (Figure 2). Two cloths are used—one square cloth on the outside and an oblong cloth for the legs, elbows, and forearms. The inner, oblong cloth is not fastened. Around the upper part of the body, the second, square cloth fits firmly (but not too tightly) in such a way that arms stay straight without being able to wriggle free but chest excursion is not hindered. From the hips downward there should be enough room within the cloths for the legs to be able to flex and abduct, allowing natural hip development. The outer cloth encloses the infant from shoulders to feet and is held together below the chin and at the height of the forearms with a safety pin. Just below the feet we secure the cloths with a short piece of string. A swaddled infant always should be put to bed with an uncovered head in a supine position. Depending on the room temperature, the sheet and the woolen, cotton, or thin fleece blanket should cover the baby up to his or her chin and be tucked tightly under the mattress. In winter, when heating is required, the room temperature preferably should be between 59° and 65°F (15°C to 18°C).

In this way, continual self-stimulation due to body movement (which tends to keep a child awake or wake him/her up too soon) is minimized. Swaddling decreases spontaneous arousal during quiet sleep and prolongs the duration of REM sleep (Gerard, Harris, & Thach, 2002b; van Sleuwen et al., 2007). Because swaddled infants are always placed in the supine sleep position, swaddling also conforms to suggestions to help prevent crib death risks associated with the prone position (Gerard, Harris, & Thach, 2002a, 2002b).

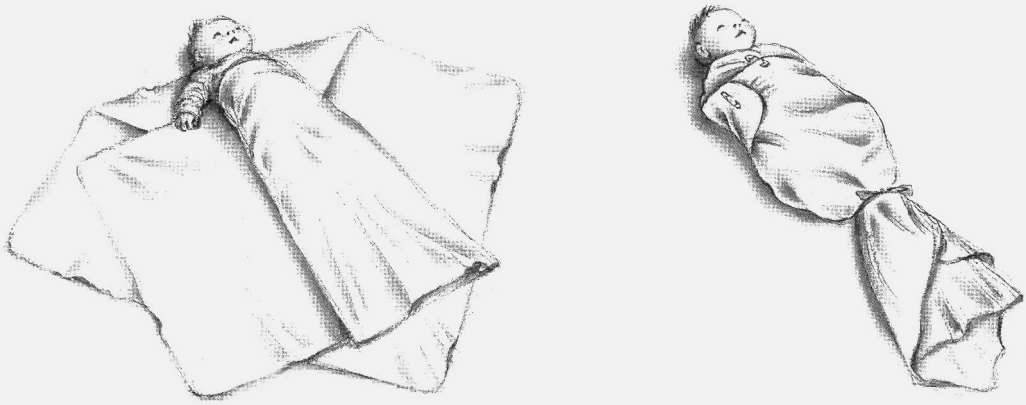


Figure 2. The swaddling method (Blom, 2005b). Drawings by Veronica Nahmias.

Contraindications to Swaddling

When illness or hunger is the cause of excessive crying, swaddling should not be applied. Other contraindications are:

- history of or an increased risk for dysplastic hip development
- gastroesophageal reflux disease
- exudative eczema
- fever or fever anticipated the first 24 hours after vaccination
- respiratory infections with violent coughing and/or fever
- reaching the age of 6 months
- attempting to turn over to the prone position.

Safe Swaddling

There are also some risks involved in swaddling an infant, and parents should be made aware of them. A safe swaddling technique should conform to the following requirements (Gerard et al., 2002b):

- safe swaddling allows leg flexion and abduction, which are necessary for a healthy hip development
- safe swaddling allows chest excursion, which is necessary for unimpeded respiration
- safe swaddling should limit the infant's ability to break free from the swaddling cloths, because there could be a risk of suffocation from loose swaddling cloths
- safe swaddling places the infant in the supine position

Nursing Contact with Parents Involved in Interventions for Excessive Crying Infants

In the Netherlands, we use a consultation process of two face-to-face discussions (60-90 minutes and 30 minutes) and three telephone consultations (15 minutes each) between nurses and parents during the intervention time period (8 weeks). In individual cases more contacts may be necessary.

First face-to-face consultation – day 1 (60-90 min)

When pediatric nurses are working with parents who have an infant who cries excessively, we recommend that they first gather data about the woman's pregnancy, birth, postnatal period, and feeding history. Physical illness or hunger as possible causes of the infant's excessive crying need to be excluded first. Subsequently, the *what, where, when,* and *how* of sleeping, feeding, and being awake have to be understood. This can be accomplished by completing a diary of a part of an average, recent day together with the parents (Appendix III shows a sample diary). Discussing this diary is often an eye-opening experience for parents, because they can see on paper that the culprits in excessive crying are actually irregularity and unpredictability in the care of the infant. The diary also demonstrates whether the infant is able to fall asleep on his or her own and how the infant's sleep deficiency and overtiredness can lead to discontentedness, inability to be or play on his or her own, restlessness, overactive behavior, and an increasing inability to fall asleep on his or her own. Next, a detailed explanation of the indicated intervention should follow. In the Netherlands, the policy is to start with the behavioral intervention of providing regularity, and if no improvement is seen after about 5 days, swaddling is added if not contraindicated. In crisis situations and when it is suspected that parents are incapable of coping with the temporary increase of crying on the first day, swaddling is started right away.

Explanation of the new approach to the parents

Advice and support for parents are directed toward helping the parents change their attitudes and their reactions to the infant's behavior and modifying their behaviors in child care. Discussion of the 24-hour diary of the infant's behavior provides a way for nurses to connect with the parents surrounding sleeping, feeding, cuddling/playing together and alone, crying, and fussing. The added column "where is your baby" indicates the degree of dependence (Blom, 2005b; Appendix III). We have found that when parents use the diary for several days, the change from chaos to regularity and from dependence to relative independence becomes visible at a glance. When swaddling is indicated, nurses should explain this technique thoroughly, including the contraindications and the risks, and help parents to swaddle the infant during the consultation. Parents should receive a written guide in which the regularity approach and, if needed, the technique of swaddling are explained (Blom 2001, 2005a).

The first two telephone consultations – day 2 and day 5 (15 minutes each)

During the Day 5 telephone call, if there has been no improvement, nurse and parents together can decide to add swaddling. In these cases we recommend that discussions about this take place in a face-to-face contact in order to fully teach about swaddling. If, after 2 to 3 days there is still no improvement the child has to be seen by a pediatrician.

Second face-to-face consultation – day 8 (30 minutes)

This consultation also has a supportive function, reviewing with the parents how the modified behavior is progressing. The 24-hour diary should be re-evaluated to determine whether regular and uniform daily infant care is being followed and its effect on the infant's behavior. If the parents are using swaddling, the nurse should ask a parent to demonstrate the way the infant is being swaddled. The nurse can expect that most infants will develop their own stable sleep-wake rhythm within 1 to 2 weeks. The possibility of a more flexible way of dealing with the infant is then discussed while upholding predictability in sleeping and drinking. Deviation from regularity, however, should be the exception rather than the rule. It is explained that to maintain the infant's recognition of being fed after waking up and being put to sleep when tired is most important.

Third telephone consultation – week 8 (15-30 minutes)

This nursing consultation is mainly intended to check whether parents, in applying and maintaining the regularity approach, have encountered difficulties related to new steps in the infant's development. If swaddling has been used, advice has to be given on how to discontinue the swaddling, which can usually be done after 6 to 8 weeks. Swaddling can be discontinued all at once by replacing it with a baby sleeping sack or as a step-by-step approach, which first leaves one arm free from swaddling. If there is no regression to excessive crying and unrest after a few sleeping periods with modified swaddling, then the second arm also can be left free. During this time, the inner cloth should remain fastened with a few safety pins so there is sufficient room for the legs to flex and abduct. In case of renewed unrest due to the freely moving arms, the infant can be swaddled again for another week, after which a new attempt to stop swaddling should be made. *When a swaddled infant makes an attempt to turn over to the prone position, swaddling must be stopped immediately.* When an infant is swaddled, it is necessary to keep in regular contact with the parents about how they are succeeding in ceasing swaddling. Swaddling should be discontinued before the age of 6 months.

Case Report Bob, 10 weeks old

First face-to-face consultation: Day 1

Bob, 10 weeks old and the second infant in the family, looks well fed. During this consultation he shouts nearly continuously and has a red complexion. His arms and legs are continually and involuntarily moving and extending. His whole body is tense. The mother reports that pregnancy and birth were uncomplicated, the Apgar score was 9/10, and birth weight was 3 kg (6 pounds, 6 ounces). In the second week he became increasingly restless. At night Bob sleeps in his own bed with one short feed, and at 6 o'clock he awakes in a good mood, but just for a while. He cannot be content on his own. During the day he catnaps in his mother's arms, the playpen, and the bouncy chair or car, and he often awakens suddenly, crying. He cries a large part of the day, even when carried.

Second face-to-face consultation: Day 8

Bob has changed completely. His eyes are calm, he no longer looks frightened, and he is not tense anymore. He is able to relax and control his own movements. He does not cry; in fact, he sometimes laughs. Ever since the second day of the intervention, Bob has developed a good sleep-feed rhythm. During the day he sleeps three periods of 1.5 to 2.5 hours in his own bed, and during the night he sleeps a period of 8.5 hours, with a short break for his bottle. Now he only cries when he is hungry and just a little before falling asleep. His mother tells that she had gotten used to reacting immediately when her infant fussed, but now she understands that her behavior stimulated her infant.

Third telephone consultation: Week 8 (15-30 minutes)
Mother: *"Bob has changed a lot, his crying a little before falling asleep also disappeared. He is able to play in the playpen alone contentedly and he is developing well. After 6 weeks we stopped swaddling, but we carried on with the regularity, because that is what makes the difference. His sleep rhythm has not changed since."*
After 1 week, a stable sleep-wake rhythm is established and Bob cries much less. It is likely that the decrease in crying after 2 weeks, which now proceeds less quickly, follows its natural course and can partly be interpreted as a normal maturational decline.

Clinical Implications

This behavioral regularity intervention, with or without swaddling, has formed the basis of a new maternal and child nursing guideline, initiated by ActiZ (Association of Health Care Providers/ActiZ, 2007), that has been provisionally approved by the Dutch National Institute for Public Health and the Environment (RIVM). The involved authors and nurses cooperated in providing nationwide information courses for nurses, physicians, and pediatricians. Regularity and uniformity in everyday childcare, presented in this article as a series of concrete suggestions (Box 1), can be taught easily by nurses to parents. Our maxim has been—and continues to be— that a contented infant means a contented parent. It is both remarkable and gratifying that the individualized nursing intervention program (the REST routine) developed by Keefe et al. (1988, 2005), which is based on a developmental psychobiological model of infant colic, resembles in many respects the regularity approach we used for our program. Although the specific ways in which the approaches are put into practice differ (REST is a homebased treatment program that emphasizes the individual needs and unique features of each family, whereas our behavioral regularity approach offers parents an easy-to-learn set of daily infant caring rules), the aim of both approaches is the same: to establish regularity and predictability in sleeping, feeding, and activity routines for the benefit of children and parents. In studies by Keefe et al. (2006) and van Sleuwen et al. (2006), parenting stress was reduced and the parents worried less.

Excessive infant crying has been shown to carry with it a serious risk of child abuse (Reijneveld et al., 2004). Because we know that behavioral regularity intervention can reduce parenting stress, it is possible that it may improve coping skills of parents, which could translate to an improvement in the overall health of the family and a possible reduction in abusive situations. More research on this is needed. Employing regularity and uniformity in daily infant care might also be a primary preventive measure. Clinical experience seems to indicate that when starting the regularity approach about 1 to 2 weeks after birth, unrest and excessive crying can be prevented (Blom, 2005b). Research is needed to confirm or refute this.

Nurses who understand behavioral methods support parents with their excessively crying infants can provide an invaluable service to the families they serve. While the healthcare systems in the Netherlands and the United States are different, modifications of the program developed and used in the study by van Sleuwen et al. (2006) and described here can be made, and nurses can adapt these interventions to match the healthcare situations in which they find themselves dealing with frustrated parents and infants who cry excessively.

References

ActiZ. (2007). Guideline Interventions for excessive crying in infants [In Dutch]. Bunnik, The Netherlands: ActiZ. www.actiz.nl

Barr, R. G., Kramer, M. S., Boisjoy, C., McVey-White, L., & Pless, I. B. (1988). Parental diary of infant cry and fuss behaviour. *Archives of Diseases in Childhood, 63,* 380-387.

Blom, R. (2001). *Rest through regularity and swaddling: A parent's guide* [in Dutch]. Utrecht, The Netherlands: De Bakermat.

Blom, R. (2005a). *Regularity gives rest: A parent's guide* [in Dutch]. Utrecht, The Netherlands: De Bakermat.

Blom, R. (2005b). *Crying and restlessness in babies: A parent's guide to natural sleeping.* Edinburg, Scotland: Floris Books.

Gerard, C. M., Harris, K. A., & Thach, B. T. (2002a). Physiologic studies on swaddling: An ancient child care practice, which may promote the supine position for infant sleep. *The Journal of Pediatrics, 141,* 398-404.

Gerard, C. M., Harris, K. A., & Thach, B. T. (2002b). Spontaneous arousals in supine infants while swaddled and unswaddled during rapid eye movement and quiet sleep. *Pediatrics, 110,* e70.

Hofacker, N., von Papoušek, M., Jacubeit, T., & Malinowski, M. (1999). Rätsel der Säuglingskoliken: Ergebnisse, Erfahrungen und therapeutische Interventionen aus der "Müncher Sprechstunde für Schreibabies.? *Monatsschrift Kinderheilkunde, 147,* 244-253.

Keefe, M. (1988). Irritable infant syndrome: Theoretical perspectives and practice implications. *Advances in Nursing Science, 10,* 70-78.

Keefe, M. R., Barbosa, G. A., Froese-Fretz, A., Kotzer, A. M., & Lobo, M. (2005). An intervention program for families with irritable infants. *MCN The American Journal of Maternal Child Nursing, 30,* 230-236.

Keefe, M. R., Karlsen, K. A., Lobo, M. L., Kotzer, A. M., & Dudley, W. N. (2006). Reducing parenting stress in families with irritable infants. *Nursing Research, 55,* 198-205.

Lucassen, P. L. B. J., Assendelft, W. J. J., Gubbels, J. W., Eijk, J. T. M., van Geldrop, W. J., van Knuistingh Neven, A. (1998). Effectiveness of treatments for infantile colic: Systematic review. *British Medical Journal, 316,* 1563-1569.

Reijneveld S. A., van der Wal, M. F., Brugman E., Hira Sing R., & Verloove- Vanhorick S. P. (2004). Infant crying and abuse. *Lancet, 364,* 1340-1342.

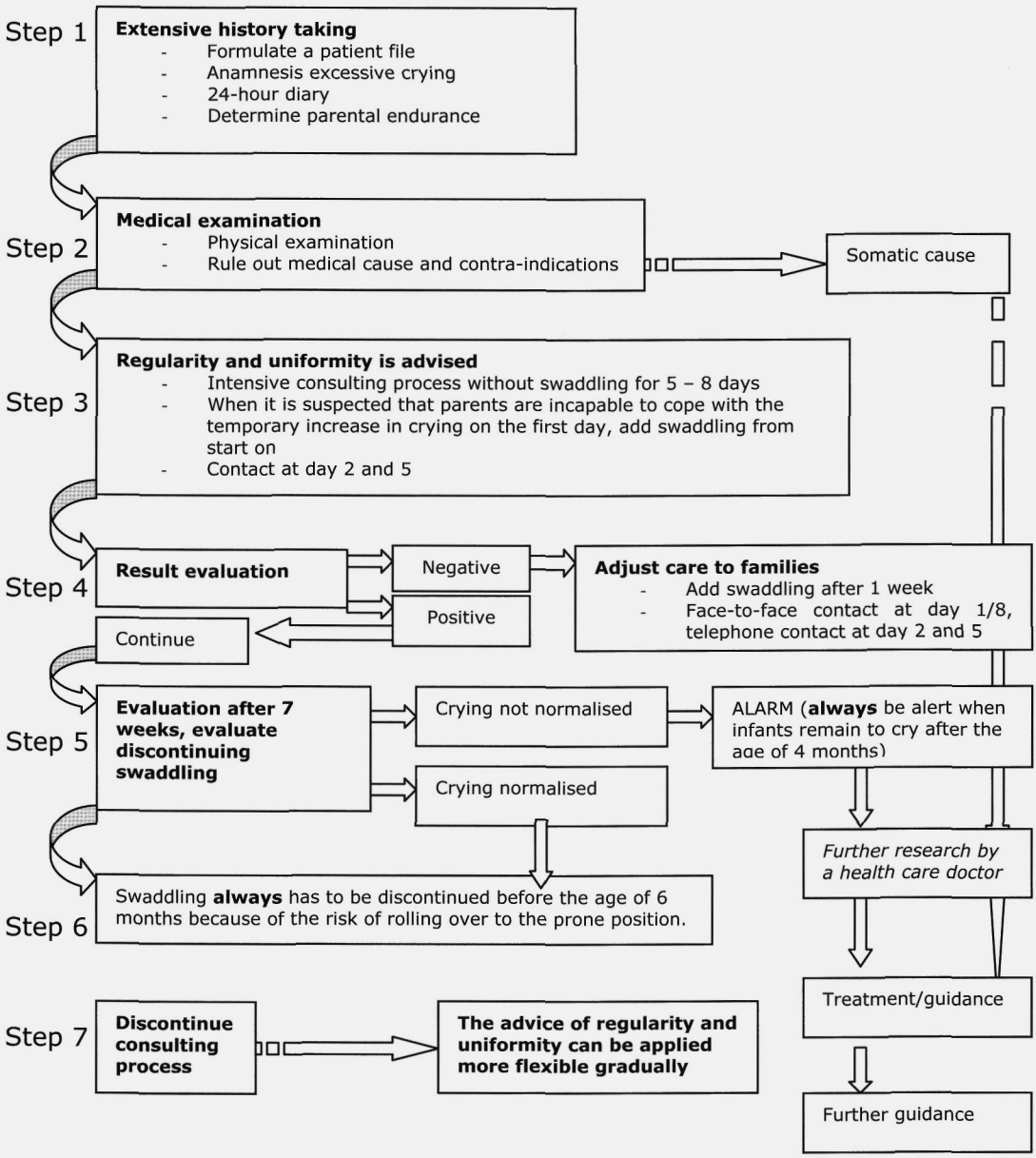
Roberts, D. M., Ostapchuk, M., & O'Brien, J. G. (2004). Infantile colic. *American Family Physician, 70,* 735-740.

van Sleuwen, B. E., L'Hoir, M. P., Engelberts, A. C., Busschers, W. B., Westers, P., Blom, M. A., et al. (2006). Comparison of behaviour modification with or without swaddling as interventions for excessive crying. *The Journal of Pediatrics, 149,* 512-517.

van Sleuwen, B. E., Engelberts, A. C., Boere-Boonekamp, M. M., Kuis, W., Schulpen, T. W. J., & L'Hoir, M. P. (2007). Swaddling: A systematic review. *Pediatrics,* 120, e1097-1106.

Appendix II

Flowchart


Step 1
Extensive history taking
- Formulate a patient file
- Anamnesis excessive crying
- 24-hour diary
- Determine parental endurance
Step 2
Medical examination
- Physical examination
- Rule out medical cause and contra-indications
Somatic cause
Step 3
Regularity and uniformity is advised
- Intensive consulting process without swaddling for 5 – 8 days
- When it is suspected that parents are incapable to cope with the temporary increase in crying on the first day, add swaddling from start on
- Contact at day 2 and 5
Step 4
Result evaluation
Negative
Positive
Adjust care to families
- Add swaddling after 1 week
- Face-to-face contact at day 1/8, telephone contact at day 2 and 5
Continue
Step 5
Evaluation after 7 weeks, evaluate discontinuing swaddling
Crying not normalised
ALARM (always be alert when infants remain to cry after the age of 4 months)
Crying normalised
Further research by a health care doctor
Step 6
Swaddling always has to be discontinued before the age of 6 months because of the risk of rolling over to the prone position.
Treatment/guidance
Step 7
Discontinue consulting process
The advice of regularity and uniformity can be applied more flexible gradually
Further guidance

Appendix III

24-hour diary

week ..., day ...	where is the baby	crying	fussing	care taking *activities*	playing together	feeding	sleeping	baby alone and content
7:00								
10								
20								
30								
40								
50								
8:00								
10								
20								
30								
40								
50								
9:00								
10								
20								
30								
40								
50								
10:00								
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11:00								
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12:00								
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20								
30								
40								
50								
13:00								
10								
20								
30								
40								
50								
14:00								
10								
20								
30								

	where is the baby	crying	fussing	care taking activities	playing together	feeding	sleeping	baby alone and content
19:00								
10								
20								
30								
40								
50								
20:00								
10								
20								
30								
40								
50								
21:00								
10								
20								
30								
40								
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22:00								
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30								
40								
50								
23:00								
10								
20								
30								
40								
50								
0:00								
10								
20								
30								
40								
50								
1:00								
10								
20								
30								
40								
50								
2:00								
10								
20								
30								

Appendix IV

Algemene vragenlijst bij inclusie in het gerandomiseerd gecontroleerd onderzoek

Geachte heer/mevrouw,

Dit boekje bevat de vragenlijst van het landelijke onderzoek naar de effectiviteit van verschillende interventies bij baby's die excessief huilen. Wij vragen u alle vragen te beantwoorden. Indien u vragen heeft, kan de wijkverpleegkundige u helpen.

Voor het invullen:

☆ Het invullen van de vragenlijst duurt ongeveer 30 minuten.

☆ De vragen zijn in de volgende groepen ingedeeld:

(1)	Persoonlijke gegevens	pagina 2 - 3
(2)	Voeding	pagina 4
(3)	Verloop van zwangerschap, bevalling en kraambed	pagina 5 - 7
(4)	Beleving van de zwangerschap, bevalling en kraambed	pagina 8 – 10
(5)	Beleving van uw huidige situatie	pagina 11
(6)	Ziekten/allergieën in uw gezin	pagina 12
(7)	Roken	pagina 13
(8)	Ziekten/allergieën bij de baby	pagina 14
(9)	Het huilen van uw baby	pagina 15
(10)	GHQ-28	pagina 16-18
(11)	Steun uit uw omgeving-lijst	pagina 19
(12)	Huilperceptieschaal week 1	pagina 20-21
(13)	Vragen over het invullen van de vragenlijst	pagina 22

De vragen in de vragenlijst kunt u beantwoorden door een kruisje te zetten in het vakje, uw antwoord te omcirkelen of uw antwoord in te vullen op de lijn. Wanneer u meerdere antwoorden mag aankruisen, staat dit onder de vraag vermeld.

Als u alle vragen hebt beantwoord, controleer dan of u niets bent vergeten. De wijkverpleegkundige zal de lijst nog eens met u nalopen.

Ik dank u nogmaals hartelijk voor uw medewerking.

Met vriendelijke groet,

drs. B.E. van Sleuwen,
projectcoördinator

dr. M.P. L'Hoir,
projectleider

PERSOONLIJKE GEGEVENS

1. Wat is de datum van de dag dat u deze vragenlijst invult? (dag/maand/jaar) ___ - ___ - _____

2. Wat is uw volledige adres?
adres _____
postcode+woonplaats _____
telefoon _____

3. Wat is de voor- en achternaam van uw baby? _____

4. Wat is de geboortedatum van uw baby? (dag/maand/jaar) ___ - ___ - _____

5. Wat was het geboortegewicht van uw baby? _____ gram

6. Wat is het huidige gewicht van uw baby? _____ gram

7. Wat was de geboortelengte van uw baby? _____ cm

8. Wat is de huidige lengte van uw baby? _____ cm

9. Wat is het geslacht van uw baby?
□ jongen
□ meisje

10. Het hoeveelste (levendgeboren) kind is dit van u?
□ 1e kind
□ 2e kind
□ 3e kind
□ 4e kind of hoger

11. Is deze baby een eenling of één van een meerling?
□ eenling
□ *één van een tweeling*
□ één van een drieling
□ één van een vier- of meerling

12. Wat is de geboortedatum van de moeder? (dag/maand/jaar) ___ - ___ - _____

13. Wat was de leeftijd van de moeder bij de geboorte van het eerste kind? _____ jaar

14. Wat is het geboorteland van de moeder? _____

15. Tot welke bevolkingsgroep rekent de moeder zich? _____

16. Indien u in het buitenland geboren bent, hoe lang bent u in Nederland? _____ jaar

17. Wat is de geboortedatum van de partner? (dag/maand/jaar) ___ - ___ - _____

18. Wat is het geboorteland van de partner? _____

19. Tot welke bevolkingsgroep rekent uw partner zich? _____

20. Indien uw partner in het buitenland geboren is, hoe lang is hij/zij in Nederland? _____ jaar

21. Bent u ....
□ ongehuwd
□ gehuwd
□ gescheiden
□ *weduwe/weduwnaar*
□ anders, nl. _____

22. Wat is uw woonsituatie?
□ samenwonend met partner en kind (-eren)
□ alleenwonend met kind (-eren)
□ anders, nl. _____

Nu volgen er een aantal vragen over opleiding en beroep. Bij de vragen over opleiding wordt gevraagd de hoogste opleiding die u of uw partner heeft afgemaakt aan te kruisen. Bij de vragen over beroep geldt huisvrouw/-man ook als beroep. Indien u werkloos of arbeidsongeschikt bent, graag het laatst uitgeoefende beroep vermelden.

23. Wat is de hoogst afgemaakte opleiding van de moeder?
- □ geen school afgemaakt
- □ lagere school
- □ (M)ULO, MAVO, Lager Beroepsonderwijs
- □ HBS, Gymnasium, Atheneum, HAVO, MMS, MBO
- □ Universiteit, HBO
- □ anders, nl. _____

24. Wat is het huidige beroep van de moeder? _____

25. Indien u buitenshuis werkt, hoeveel uur werkt u dan gemiddeld per week? _____ uur

26. Wat is de hoogst afgemaakte opleiding van uw partner?
- □ geen school afgemaakt
- □ lagere school
- □ (M)ULO, MAVO, Lager Beroepsonderwijs
- □ HBS, Gymnasium, Atheneum, HAVO, MMS, MBO
- □ Universiteit, HBO
- □ anders, nl. _____

27. Wat is het huidige beroep van uw partner? _____

28. Indien de partner buitenshuis werkt, hoeveel uur werkt hij/zij dan gemiddeld per week?
_____ uur

VOEDING

29. Hoe wordt uw baby op dit moment gevoed? (deze vraag gaat alleen over fles- of borstvoeding, bijvoeding komt later aan de orde)
- ☐ alleen borstvoeding
- ☐ alleen kunst(fles-)voeding
- ☐ zowel borst- als kunst(fles-)voeding
- ☐ kunst(fles-)- en papvoeding
- ☐ anders, nl. _____

30. Hoe lang heeft uw baby vanaf de geboorte uitsluitend borstvoeding gekregen? _____ weken

31. Als uw baby kunst(fles-)voeding krijgt, welke voeding is dat momenteel? _____

32. Hoeveel maal per 24 uur geeft u uw baby borstvoeding? _____ borstvoedingen per 24 uur

33. Hoeveel maal per 24 uur geeft u uw baby kunst(fles-)voedingen?
_____ kunst(fles-)voedingen per 24 uur

34. Hoeveel voeding geeft u gemiddeld per keer in een fles?
_____ ml gemiddeld per voeding

35. Heeft uw baby voor de huidige kunst(fles-)voeding andere soorten kunst(fles-)voeding gebruikt?
- ☐ ja, nl. _____
- ☐ nee

36. Hoeveel papvoedingen gebruikt uw baby per 24 uur? _____ papvoedingen per 24 uur

37. Wat gebruikt u meestal om de papvoeding van te maken?
- ☐ gewone zuigelingenvoeding
- ☐ opvolgmelk
- ☐ halfvolle melk
- ☐ volle melk
- ☐ anders, nl. _____

VERLOOP VAN ZWANGERSCHAP, BEVALLING EN KRAAMBED

38. Is uw zwangerschap normaal verlopen? □ ja (ga naar vraag 40)
□ nee

39. Zo nee, waarom is deze niet normaal verlopen? □ kind groeide slecht
(U kunt meerdere mogelijkheden aankruisen) □ te vroege weeënactiviteit
□ zwanger van een twee- of meerling
□ abnormaal bloedverlies, daardoor ziekenhuisopname
□ hoge bloeddruk
□ nauw bekken
□ verkeerde ligging van het kind
□ anders, nl. _____

40. Had u de volgende klachten tijdens de zwangerschap?
(Zet een kruisje in het vakje onder ja of nee)

Klachten tijdens zwangerschap	ja	nee
Moeheid		
Zuurbranden		
Misselijkheid		
Steken in de borst		
Opgeblazen gevoel		
Constipatie/verstopping		
Spataderen		
Bekkeninstabiliteit		
Krampen/harde buiken		
Overige klachten:		

41. Had u de volgende symptomen van ziekte tijdens de zwangerschap?
(Zet een kruisje in het vakje onder ja of nee)

Symptomen van ziekte	ja	nee
Migraine		
Hoofdpijn		
Rugpijn		
Opgezette handen en voeten		
Huidklachten		
Overige symptomen:		

42. Hoe schat u uw eigen gezondheid in tijdens de zwangerschap? (één cijfer omcirkelen)

1	2	3	4	5	6	7
goed						slecht

43. Lijdt u aan één van de volgende ziekten?
(Zet een kruisje in het vakje onder ja of nee)

Ziekten	ja	nee
Diabetes		
Epilepsie		
Verlaagde schildklierfunctie		
Verhoogde schildklierfunctie		
Hoge bloeddruk		
Allergie(en)		
Reuma		
Coeliakie		
Psychische klachten/problemen		
Terugkerende urineweginfecties		
Rugklachten		
Overige klachten/problemen:		

44. Hoeveel dagen was u tijdens uw zwangerschap afwezig van uw werk? _____ dagen

45. Vond u uw werk tijdens uw zwangerschap vermoeiender dan daarvoor?
- □ ja
- □ enigszins
- □ nee

46. Hoeveel weken was u zwanger toen de baby geboren werd? _____ weken

47. Hoe lang duurde uw bevalling in totaal? (in uren) _____ uur

48. Hoe lang heeft u persweeën gehad? (in minuten) _____ minuten

49. Waar vond de bevalling plaats?
- □ thuis
- □ gedeeltelijk thuis, gedeeltelijk in het ziekenhuis
- □ poliklinisch in het ziekenhuis
- □ op medische indicatie in het ziekenhuis
- □ anders, nl. _____

50. Als u in het ziekenhuis onder leiding van een gynaecoloog bent bevallen, wat was hiervoor de reden? (u kunt meerdere mogelijkheden aankruisen)
- □ ontsluitingsweeën duurden te lang
- □ persen duurde te lang
- □ keizersnede
- □ tangverlossing
- □ zuignap-/vacuumverlossing
- □ anders, nl. _____

51.Heeft uw baby in de couveuse gelegen?
- □ ja
- □ nee (ga naar vraag 53)

52. Zo ja, wat was hiervoor de reden? _____

53. Had uw baby na de geboorte geboorteletsel?
- □ ja
- □ nee (ga naar vraag 55)

54. Zo ja, wat voor geboorteletsel was dit? (u kunt meerdere mogelijkheden aankruisen)
- □ sleutelbeenbreuk
- □ het armpje bewoog niet goed
- □ vervormd hoofd
- □ anders, nl. _____

55. Wat was de APGAR-score na 1 minuut? _____

56. Wat was de APGAR-score na 5 minuten? _____

57. De kraamperiode heb ik:
- ◻ geheel thuis doorgebracht
- ◻ deels thuis, deels in het ziekenhuis doorgebracht
- ◻ geheel in het ziekenhuis doorgebracht
- ◻ anders, nl. _____

58. Is het kraambed normaal verlopen?
- ◻ ja (*ga naar vraag 60*)
- ◻ nee

59. Zo nee, waarom is dit niet normaal verlopen? (U kunt meerdere mogelijkheden aankruisen)
- ◻ hevig bloedverlies, daardoor ziekenhuisopname
- ◻ borstontsteking
- ◻ *baarmoederontsteking*
- ◻ trombosebeen
- ◻ anders, nl. _____

BELEVING VAN DE ZWANGERSCHAP, BEVALLING EN KRAAMBED

Bij de volgende vragen kunt u steeds één cijfer omcirkelen. Als u bijvoorbeeld vond dat uw zwangerschap niet makkelijk, maar toch ook niet moeilijk was, dan zet u een rondje om cijfer 3.

60. Hoe heeft u uw zwangerschap ervaren?

zeer positief	1	2	3	4	5	zeer negatief

61. Hoe heeft uw partner uw zwangerschap ervaren?

zeer positief	1	2	3	4	5	zeer negatief

62. De zwangerschap was....

makkelijk	1	2	3	4	5	moeilijk

63. Is de zwangerschap volgens uw verwachting verlopen?

ja, volledig	1	2	3	4	5	nee, helemaal niet

64. De zwangerschap is....

meegevallen	1	2	3	4	5	tegengevallen

65. Kon uw partner zich verplaatsen in uw gemoedstoestand tijdens de zwangerschap?

ja, volledig	1	2	3	4	5	nee, helemaal niet

66. Begreep uw partner uw wijze van denken tijdens de zwangerschap?

ja, volledig	1	2	3	4	5	nee, helemaal niet

67. Veranderde uw relatie tijdens de zwangerschap?

werd beter	1	2	3	4	5	verslechterde

68. Veranderde uw seksuele relatie tijdens de zwangerschap?

werd beter	1	2	3	4	5	verslechterde

69. Had u het gevoel overal alleen voor te staan tijdens de zwangerschap?

nee, helemaal niet	1	2	3	4	5	ja, volledig

70. Had u veel emotionele steun tijdens de zwangerschap?

ja, veel steun	1	2	3	4	5	nee, geen steun

71. Voelde u zich geïsoleerd van anderen tijdens de zwangerschap?

nee, nooit	1	2	3	4	5	ja, vaak

72. Had u tijdens de zwangerschap minder contact met vrienden dan gewoonlijk?

nee, evenveel	1	2	3	4	5	ja, veel minder

73. Hoe heeft u de bevalling ervaren?

zeer positief	1	2	3	4	5	zeer negatief

74. Hoe heeft uw partner de bevalling ervaren?

zeer positief	1	2	3	4	5	zeer negatief

75. De bevalling was....

makkelijk	1	2	3	4	5	moeilijk

76. De bevalling is ....

meegevallen	1	2	3	4	5	tegengevallen

77. Bent u teleurgesteld in uw vrouw-zijn?

nee, helemaal niet	1	2	3	4	5	ja, heel erg

78. Het kraambed was ....

makkelijk	1	2	3	4	5	moeilijk

79. Het kraambed is ....

meegevallen	1	2	3	4	5	tegengevallen

80. Voelde u zich onzeker na de geboorte van uw baby?

nee, helemaal niet	1	2	3	4	5	ja, heel erg

81. Had u de eerste paar weken na de bevalling sterke behoefte aan hulp van anderen?

nee, geen behoefte	1	2	3	4	5	ja, veel behoefte

82. Durft u de zorg voor uw baby aan anderen over te laten?

ja, vaak	1	2	3	4	5	nee, nooit

83. Voelt u zich sinds de geboorte in uw vrijheid beperkt?

nee, helemaal niet	1	2	3	4	5	ja, sterk beperkt

84. Hoe vaak per jaar gaat u gewoonlijk naar:
(geef een schatting van het gemiddeld aantal keer per jaar)

de bioscoop	_____ keer per jaar
het theater	_____ keer per jaar
een concert	_____ keer per jaar
een (sport)wedstrijd	_____ keer per jaar
de bibliotheek	_____ keer per jaar
een lezing/voordracht	_____ keer per jaar

De volgende vragen gaan over uw gedachten in de eerste drie maanden van uw zwangerschap. Probeert u zich in te leven in die periode en de vragen zo goed mogelijk te beantwoorden. U kunt het antwoord wat het meest op u van toepassing is omcirkelen.

1 absoluut niet van toepassing
2 nauwelijks van toepassing
3 soms
4 redelijk van toepassing
5 zeer goed van toepassing

	1	2	3	4	5
85. Ik was in de eerste drie maanden van mijn zwangerschap bang voor pijn bij de weeën en de bevalling.	□	□	□	□	□
86. Ik was in de eerste drie maanden van mijn zwangerschap bang voor de bevalling omdat ik er nog nooit een had meegemaakt.	□	□	□	□	□
87. Ik was er in de eerste drie maanden van mijn zwangerschap bang voor dat ik me niet zou kunnen beheersen tijdens de bevalling en zou schreeuwen.	□	□	□	□	□
88. Ik was in de eerste drie maanden van mijn zwangerschap bang dat mijn baby geestelijk gehandicapt zou zijn of hersenletsel zou hebben.	□	□	□	□	□
89. Ik was in de eerste drie maanden van mijn zwangerschap bang dat mijn baby dood geboren zou worden, of tijdens of direct na de geboorte zou sterven.	□	□	□	□	□
90. Ik was in de eerste drie maanden van mijn zwangerschap bang dat mijn baby een lichamelijk gebrek zou hebben of lichamelijk niet helemaal in orde zou zijn.	□	□	□	□	□
91. Ik dacht er in de eerste drie maanden van de zwangerschap wel eens aan dat mijn baby lichamelijk een ziekelijk of zwak kindje zou worden.	□	□	□	□	□

Bron: dr. A.C. Huizink, Universiteit Utrecht. Proefschrift. Pregnancy Related Anxiety Questionnaire-Revised.

BELEVING VAN UW HUIDIGE SITUATIE

Op deze bladzijde vindt u een lijst met woorden. Deze woorden beschrijven gevoelstoestanden. Het is *de bedoeling dat u aangeeft in welke mate de betekenis van het woord past bij uw gemoedstoestand* **op dit moment**.

Bijvoorbeeld: "prettig" 0 1 2 (3) 4

Als het woord **absoluut niet** bij uw gevoel past, dus als u zich helemaal niet prettig voelt, dan omcirkelt u het cijfer 0.
Als het woord **een beetje** bij uw gevoel past, dus als u zich weinig prettig voelt, dan omcirkelt u het cijfer 1.
Als het woord **middelmatig** bij uw gevoel past, dus als u zich prettig voelt, dan omcirkelt u het cijfer 2.
Als het woord **goed** *bij uw gevoel past, dus als u zich erg prettig voelt, dan omcirkelt u het cijfer* 3.
Als het woord **heel goed** bij uw gevoel past, dus als u zich bijzonder prettig voelt, dan omcirkelt u het cijfer 4.

Het gaat er dus om hoe u zich op het moment voelt.

Denk niet te lang na over uw antwoord. Het gaat om uw eerste indruk. Er bestaan geen foute antwoorden. Elk antwoord is goed, als het uw eigen stemming weergeeft. Sla geen woorden over.

De omschrijving past bij mijn gevoel van dit moment.

0 = absoluut niet 1 = een beetje 2 = middelmatig 3 = goed 4 = heel goed

1.	neerslachtig	0	1	2	3	4
2.	slecht gehumeurd	0	1	2	3	4
3.	uitgeput	0	1	2	3	4
4.	actief	0	1	2	3	4
5.	zenuwachtig	0	1	2	3	4
6.	hulpeloos	0	1	2	3	4
7.	geërgerd	0	1	2	3	4
8.	helder	0	1	2	3	4
9.	paniekerig	0	1	2	3	4
10.	droevig	0	1	2	3	4
11.	opstandig	0	1	2	3	4
12.	vermoeid	0	1	2	3	4
13.	levendig	0	1	2	3	4
14.	gespannen	0	1	2	3	4
15.	eenzaam	0	1	2	3	4
16.	aan het eind van mijn krachten	0	1	2	3	4
17.	ongelukkig	0	1	2	3	4
18.	woedend	0	1	2	3	4
19.	lusteloos	0	1	2	3	4
20.	vol energie	0	1	2	3	4
21.	rusteloos	0	1	2	3	4
22.	onwaardig	0	1	2	3	4
23.	knorrig	0	1	2	3	4
24.	doodop	0	1	2	3	4
25.	opgeruimd	0	1	2	3	4
26.	angstig	0	1	2	3	4
27.	droefgeestig	0	1	2	3	4
28.	kwaad	0	1	2	3	4
29.	afgemat	0	1	2	3	4
30.	onzeker	0	1	2	3	4
31.	wanhopig	0	1	2	3	4
32.	mopperig	0	1	2	3	4

ZIEKTEN/ALLERGIEEN IN UW GEZIN

92. Heeft u ooit last gehad van:
(Zet een kruisje in het vakje onder ja of nee)

	ja	nee
Eczeem (als kind) (behalve zwemmerseczeem)		
Hooikoorts		
Astma of bronchitis		
Voedselallergie		

93. Heeft de vader van de baby ooit last gehad van:
(Zet een kruisje in het vakje onder ja of nee)

	ja	neë
Eczeem (als kind) (behalve zwemmerseczeem)		
Hooikoorts		
Astma of bronchitis		
Voedselallergie		

94. Gebruikte u medicijnen of kalmerende middelen tijdens uw zwangerschap?
(met uitzondering van vitamine en/of ijzerpreparaten) □ ja
□ nee (ga naar vraag 96)

95. Zo ja, welke? _____ waarvoor? _____
_____ waarvoor? _____
_____ waarvoor? _____

96. Gebruikt u op dit moment medicijnen of kalmerende middelen □ ja
(met uitzondering van vitamine en/of ijzerpreparaten) □ nee (ga naar vraag 98)

97. Zo ja, welke? _____ waarvoor? _____
_____ waarvoor? _____
_____ waarvoor? _____

98. Is in uw familie ooit wiegendood voorgekomen? □ ja
□ nee

99. Zijn er in uw familie wervelkolomproblemen bekend? □ ja
(bv. scoliose, geboorteafwijkingen, beenverkorting) □ nee

100. Hebben u en/of uw partner regelmatig nek- en lage rugklachten? □ ja
(bv. nek-, hoofdpijn, migraine, hernia) □ nee

101. Werden kinderen uit uw gezin ooit behandeld voor houdingsproblemen?
□ ja
□ nee

ROKEN

102. Heeft u gerookt tijdens uw zwangerschap?
- ☐ ja
- ☐ nee (ga naar vraag 104)

103. Zo ja, hoeveel sigaretten/shag rookte u toen gemiddeld per 24 uur?
- ☐ 1-10 sigaretten/shag per 24 uur
- ☐ 11-20 sigaretten/shag per 24 uur
- ☐ 21-30 sigaretten/shag per 24 uur
- ☐ 31 of meer sigaretten/shag per 24 uur

104. Rookt u nu?
- ☐ ja
- ☐ nee (ga naar vraag 107)

105. Zo ja, rookt u ook **in huis**?
- ☐ ja
- ☐ nee (ga naar vraag 107)

106. Zo ja, hoeveel sigaretten/shag rookt u nu gemiddeld per 24 uur **in huis**?
- ☐ 1-10 sigaretten/shag per 24 uur
- ☐ 11-20 sigaretten/shag per 24 uur
- ☐ 21-30 sigaretten/shag per 24 uur
- ☐ 31 of meer sigaretten/shag per 24 uur

107. Rookt uw partner ook **in huis**?
- ☐ ja
- ☐ nee (ga naar vraag 109)

108. Zo ja, hoeveel sigaretten/shag rookt hij/zij gemiddeld per 24 uur **in huis**?
- ☐ 1-10 sigaretten/shag per 24 uur
- ☐ 11-20 sigaretten/shag per 24 uur
- ☐ 21-30 sigaretten/shag per 24 uur
- ☐ 31 of meer sigaretten/shag per 24 uur

109. Hoeveel dagen per week wordt er **in huis** gerookt? gemiddeld _____ dagen per week

ZIEKTEN/ALLERGIEEN BIJ DE BABY

110. Is uw baby sinds de geboorte ziek geweest? ☐ ja
☐ nee (ga naar vraag 112)

111. Zo ja, wat was de reden? _____
_____

112. Is uw baby sinds de geboorte opgenomen geweest in een ziekenhuis?
☐ ja
☐ nee (ga naar vraag 114)

113. Zo ja, wat was de reden? _____
_____

114. Kunt u aangeven hoe vaak het betreffende verschijnsel bij uw baby voorkomt? (s.v.p. 1 kruisje in de betreffende rij plaatsen)

	nooit	minder dan 1 maal per week	1 maal per week	1-2 maal daags	altijd
a) Heeft uw baby last van spugen of braken vlak na het voeden? (niet bedoeld wordt het teruggeven van kleine beetjes voeding)					
b) Heeft uw baby last van diarree?					
c) Heeft uw baby last van verstoppingen?					
d) Heeft uw baby last van eczeem of dauwworm?					
e) Heeft uw baby last van piepende ademhaling?					
f) Krijgt uw baby rode vlekken in het gezicht vlak na het voeden?					
g) Krijgt uw baby opgezette oogleden of lippen vlak na het voeden?					
h) Krijgt uw baby vlak na de voeding bulten op de huid die lijken op de plekken die ontstaan na contact met brandnetels?					

115. Wat viel u op bij uw baby vanaf de geboorte?
(Zet een kruisje in het vakje onder ja of nee)

	ja	nee
a) Mijn baby is (was) overstrekt		
b) Mijn baby heeft (had) een scheve houding		
c) Mijn baby heeft (had) een verkeerde voetenstand		
d) Mijn baby heeft (had) een kale plek op het achterhoofd niet in het midden		
e) Mijn baby kan (kon) niet op zijn/haar buik liggen (in de box)		
f) Mijn baby slaapt (sliep) goed in		
g) Mijn baby slaapt (sliep) goed door		
h) Het (borst-)voeden gaat (ging) altijd moeilijk aan 1 kant		

HET HUILEN VAN UW BABY

116. Kunt u aangeven hoe vaak de genoemde symptomen bij uw baby voorkomen? (s.v.p. 1 kruisje in het betreffende hokje plaatsen)

	nooit	soms	meestal	vaak
Mijn baby heeft buikpijn tijdens het huilen				
Mijn baby heeft moeite met ontlasting				
Mijn baby is te troosten als hij/zij huilt				
Mijn baby laat winden tijdens of na het huilen				
Mijn baby loopt rood aan tijdens het huilen				
Mijn baby heeft een opgezette buik tijdens het huilen				

117. Wie heeft u geraadpleegd over het huilen van uw baby? (u kunt meerdere mogelijkheden aankruisen)
- ☐ huisarts
- ☐ consultatiebureau-arts
- ☐ wijkverpleegkundige
- ☐ kinderarts
- ☐ fysiotherapeut
- ☐ homeopaat
- ☐ osteopaat
- ☐ chiropractor
- ☐ anders, nl. _____

118. Had u eerder een kind dat veel huilde? kinderrij)
- ☐ ja, mijn _____ kind (positie in de
- ☐ nee

119. Ik denk dat mijn baby ongeveer _____ uur per 24 uur huilt

120. Heeft u het gevoel dat dit kind een kwetsbaar kind is?

1 2 3 4 5 6 7
ja, absoluut nee, absoluut niet

121. Vergeleken met andere baby's huilt mijn baby veel.

1 2 3 4 5 6 7
ja, absoluut nee, helemaal niet

122. Het huilen van mijn baby is voor mij ....

1 2 3 4 5 6 7
een zeer groot probleem helemaal geen probleem

123. Vergeleken met andere baby's vind ik mijn baby ....

1 2 3 4 5 6 7
heel moeilijk heel gemakkelijk

124. Ik heb het gevoel er helemaal alleen voor te staan.

1 2 3 4 5 6 7
ja, heel erg nee, helemaal niet

GHQ – 28

Wij willen graag weten hoe gezond u zich voelt en welke klachten u DE AFGELOPEN PAAR WEKEN heeft gehad.
Wilt u daarom ALLE onderstaande vragen beantwoorden door bij iedere vraag het antwoord dat het meest op u van toepassing is te omcirkelen. Denk erom dat het bij deze vragen uitsluitend gaat om de klachten VAN DIT MOMENT OF VAN DE AFGELOPEN PAAR WEKEN en dus niet om klachten die u in het verleden ooit heeft gehad.

Bijvoorbeeld:

a1. Heeft u zich de laatste tijd helemaal fit en gezond gevoeld?	Beter dan gewoonlijk	Net zoals gewoonlijk	Slechter dan gewoonlijk	Veel slechter dan gewoonlijk

Speciale uitgave van het Wilhelmina Kinderziekenhuis te Utrecht, vervaardigd na schriftelijke toestemming dd. 5 december 2000, van de uitgever Swets en Zeitlinger B.V. te Lisse.

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A1. Heeft u zich de laatste tijd helemaal fit en gezond gevoeld?	Beter dan gewoonlijk	Net zoals gewoonlijk	Slechter dan gewoonlijk	Veel slechter dan gewoonlijk
A2. Heeft u de laatste tijd behoefte gehad aan een versterkend middel (tonicum)?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
A3. Heeft u zich de laatste tijd moe en uit uw doen gevoeld?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
A4. Heeft u de laatste tijd het gevoel Gehad dat u ziek bent?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
A5. Heeft u de laatste tijd pijn in uw hoofd gehad?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
A6. Heeft u de laatste tijd een gespannen of drukkend gevoel uw hoofd gehad?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
A7. Heeft u het de laatste tijd weleens plotseling warm of koud? gehad?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
B1. Bent u de laatste tijd door zorgen veel slaap tekort gekomen?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
B2. Heeft u de laatste tijd moeite gehad met doorslapen, nadat u eenmaal ingeslapen was?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
B3. Heeft u de laatste tijd het gevoel gehad dat u voortdurend onder druk stond?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
B4. Bent u de laatste tijd prikkelbaar en slecht gehumeurd geweest?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
B5. Bent u de laatste tijd wel eens zomaar bang geworden of in paniek geraakt?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
B6. Heeft u de laatste tijd het gevoel gehad dat alles u teveel aan het worden is?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
B7. Bent u de laatste tijd voortdurend zenuwachtig en gespannen geweest?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
C1. Heeft u zich de laatste tijd goed kunnen bezighouden?	Beter dan gewoonlijk	Net zo goed als gewoonlijk	Minder goed dan gewoonlijk	Veel minder goed dan gewoonlijk

C2. Bent u de laatste tijd langer over uw dagelijkse bezigheden gaan doen?	Sneller dan gewoonlijk	Net zo lang als gewoonlijk	Langer dan gewoonlijk	Veel langer dan gewoonlijk
C3. Heeft u de laatste tijd het gevoel dat u de dingen over het geheel genomen goed doet?	Beter dan gewoonlijk	Ongeveer zo goed als gewoonlijk	Minder goed dan gewoonlijk	Veel minder goed dan gewoonlijk
C4. Bent u de laatste tijd tevreden over de manier waarop u uw taken uitvoert?	Meer tevreden dan gewoonlijk	Ongeveer net zo tevreden als gewoonlijk	Minder tevreden dan gewoonlijk	Veel minder tevreden dan gewoonlijk
C5. Heeft u de laatste tijd het gevoel gehad zinvol bezig te zijn?	Zinvoller dan gewoonlijk	Net zo zinvol als gewoonlijk	Minder zinvol dan gewoonlijk	Veel minder zinvol dan gewoonlijk
C6. Voelde u zich de laatste tijd in staat om beslissingen (over dingen) te nemen?	Beter in staat dan gewoonlijk	Net zo goed in staat als gewoonlijk	Wat minder goed in staat dan gewoonlijk	Veel minder goed in staat dan gewoonlijk
C7. Heeft u de laatste tijd plezier kunnen beleven aan uw gewone, dagelijkse bezigheden?	Meer dan gewoonlijk	Evenveel als gewoonlijk	Wat minder dan gewoonlijk	Veel minder dan gewoonlijk
D1. Heeft u zich de laatste tijd als een waardeloos iemand beschouwd?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
D2. Heeft u de laatste tijd het gevoel gehad dat uw leven totaal uitzichtloos is?	Helemaal niet uitzichtloos	Niet uitzicht-lozer dan gewoonlijk	Wat meer uitzichtloos dan gewoonlijk	Veel meer uitzichtloos dan gewoonlijk
D3. Heeft u de laatste tijd het gevoel gehad dat het leven niet de moeite waard is?	Nee, heb dat gevoel gehad	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
D4. Heeft u de laatste tijd over-wogen om een einde aan uw leven te maken?	Zeer zeker niet	Ik dacht van niet	De gedachte is wel eens bij me opgekomen	Serieus overwogen
D5. Bent u de laatste tijd wel eens zenuwachtig geweest dat u niets meer kon doen?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
D6. Heeft u zich de laatste tijd betrapt op de gedachte: 'was ik maar dood en van alles af'?	Helemaal niet	Niet meer dan gewoonlijk	Wat meer dan gewoonlijk	Veel meer dan gewoonlijk
D7. Drong de gedachte er een eind aan te maken zich steeds weer bij u op?	Zeer zeker niet	Ik dacht van niet	De gedachte is wel eens bij mij opgekomen	Drong zich beslist aan mij op

STEUN UIT UW OMGEVING-LIJST

Bij de volgende vragen gaat het erom in welke mate het gedrag, de reactie van mensen waar u mee omgaat, afwijkt van wat u zou wensen. Het is de bedoeling dat u bij de vragen denkt aan alle mensen waar u mee omgaat (dus het geheel van familieleden, vrienden, kennissen, buren, collega's etcetera). Wilt u een kruisje zetten in het hokje dat het best bij u past.

1 mis ik, zou ik graag meer willen
2 mis ik niet echt, maar het zou prettig zijn als het iets vaker gebeurde
3 precies goed zo; ik zou niet vaker of minder vaak willen
4 gebeurt te vaak; het zou prettig zijn als het minder vaak gebeurde

		1	**2**	**3**	**4**
1	u aanhalen	□	□	□	□
2	u om raad vragen	□	□	□	□
3	u een ruggesteuntje geven	□	□	□	□
4	u laten merken wat er van u verwacht wordt	□	□	□	□
5	u ergens heen brengen	□	□	□	□
6	u opmonteren/opvrolijken	□	□	□	□
7	u knuffels/liefkozingen geven	□	□	□	□
8	u een luisterend oor bieden	□	□	□	□
9	u vragen ergens aan mee te doen	□	□	□	□
10	u een duwtje in de goede richting geven	□	□	□	□
11	u goede raad geven	□	□	□	□
12	aan u spulletjes of een klein bedrag lenen	□	□	□	□
13	u zomaar opbellen of een praatje met u maken	□	□	□	□
14	u complimenten geven	□	□	□	□
15	u in vertrouwen nemen	□	□	□	□
16	u om hulp vragen	□	□	□	□
17	u zeggen dat u moet volhouden	□	□	□	□
18	u informatie geven over waar u iets kunt krijgen	□	□	□	□
19	gezellig bij u op bezoek komen	□	□	□	□
20	u hulp bieden in bijzondere gevallen, zoals bij ziekte, verhuizing, kinderen uitbesteden	□	□	□	□
21	aan u grote dingen zoals een auto of een groot bedrag lenen	□	□	□	□
22	u advies geven bij allerlei huishoudelijke probleempjes	□	□	□	□
23	samen met u gaan winkelen, naar een film of wedstrijd gaan, of zomaar een dagje uit gaan	□	□	□	□
24	genegenheid voor u tonen	□	□	□	□
25	u opbouwende kritiek geven	□	□	□	□
26	u troosten	□	□	□	□
27	u laten begrijpen waarom u iets niet goed deed	□	□	□	□
28	uw advies opvolgen	□	□	□	□
29	u helpen uw problemen te verhelderen	□	□	□	□
30	uw sterke punten naar voren halen	□	□	□	□
31	u informatie over uw gedrag geven	□	□	□	□
32	u praktisch hulp bieden bij alledaagse dingen zoals in het huishouden of bij een klusje	□	□	□	□
33	u uitnodigen voor een feestje of etentje	□	□	□	□
34	u geruststellen	□	□	□	□

Bron: Noordelijk Centrum voor Gezondheidsvraagstukken, Rijksuniversiteit Groningen. Dr. F.L.P. van Sonderen.

HUILPERCEPTIESCHAAL

week 1

1. Hoe klinkt het huilen van uw kind?

Stel u voor dat uw baby huilt. Hoe zou u het huilen van uw baby beoordelen? Omcirkel het cijfer wat het meest op u van toepassing is.

a. Het huilen van mijn kind klinkt:

1	2	3	4	5	6	7
niet dringend						dringend

b. Het huilen van mijn kind klinkt:

1	2	3	4	5	6	7
prettig						onaangenaam

c. Het huilen van mijn kind klinkt:

1	2	3	4	5	6	7
gezond						ziek

d. Het huilen van mijn kind klinkt:

1	2	3	4	5	6	7
gekalmeerd						geprikkeld

e. Het huilen van mijn kind klinkt:

1	2	3	4	5	6	7
niet doordringend						doordringend

f. Het huilen van mijn kind klinkt:

1	2	3	4	5	6	7
opbeurend						ongemakkelijk

g. Het huilen van mijn kind:

1	2	3	4	5	6	7
staat mij niet tegen						*staat mij tegen*

h. Het huilen van mijn kind klinkt:

1	2	3	4	5	6	7
niet verontrustend						verontrustend

2. Hoe voel ik me bij het huilen van mijn baby?

Stel u voor dat uw baby huilt. Omcirkel het cijfer wat het beste uw gevoel weergeeft ten opzichte van het huilen van uw baby.

a. Hoe geïrriteerd/boos voel ik me door het huilen van mijn baby?

1 2 3 4 5 6 7

helemaal niet geïrriteerd/boos gemiddeld geïrriteerd/boos extreem geïrriteerd/boos

b. Hoe verdrietig maakt het huilen van uw baby u?

1 2 3 4 5 6 7

helemaal niet verdrietig gemiddeld verdrietig extreem verdrietig

c. Hoe verwend klinkt het huilen van uw kind?

1 2 3 4 5 6 7

extreem verwend gemiddeld verwend helemaal niet verwend

d. Geeft het huilen van uw kind u het gevoel dat u voor hem/haar wilt zorgen?

1 2 3 4 5 6 7

helemaal niet een beetje absoluut

3. Wat is de reden van het huilen van uw baby?

Wat is meestal de reden dat uw baby huilt? Geef uw eerste, tweede en derde reden aan door achter de mogelijke redenen een 1, 2 of 3 te zetten.

uw baby huilt meestal doordat....	Uw keuze (1, 2 of 3)
hij/zij een natte/vieze luier heeft	
hij/zij moe is	
hij/zij darmkrampjes heeft	
hij/zij aandacht wil krijgen	
hij/zij tandjes krijgt	
hij/zij honger heeft	
hij/zij het te koud/warm heeft	
hij/zij pijn heeft	
hij/zij ziek is	
hij/zij bang is	
hij/zij gefrustreerd is	
iets anders, nl.	

VRAGEN OVER HET INVULLEN VAN DE VRAGENLIJST

125. Hoe vond u het invullen van deze vragenlijst?

- ◻ gemakkelijk
- ◻ niet moeilijk, niet makkelijk
- ◻ moeilijk

126. Wilt u bericht ontvangen over de resultaten van dit onderzoek?

- ◻ ja, graag
- ◻ nee, liever niet

127. Mogen wij u later nog eens benaderen voor een vervolgonderzoek of aanvullende vragen?

- ◻ ja
- ◻ nee (ga naar vraag 129)

128. Zo ja, zou u uw e-mailadres en/of telefoonnummer van uw buren willen noteren, voor het geval dat u verhuist?

_____

_____

_____

129. Bij het volgende gesprek krijgt u te horen in welke onderzoeksgroep u bent ingedeeld. Heeft u zelf een voorkeur?

- ◻ ik zou graag ritme-adviezen krijgen
- ◻ ik zou graag ritme-adviezen willen krijgen en daarbij willen leren inbakeren
- ◻ ik heb geen voorkeur

Wilt u zorgvuldig controleren of alle vragen ingevuld zijn? De wijkverpleegkundige zal deze vragenlijst nog eens met u doornemen. Als er onduidelijkheden zijn, kunt u die met haar bespreken.

Tot slot willen wij u bedanken voor uw bereidheid om mee te werken. Nogmaals willen wij duidelijk stellen dat uw gegevens anoniem verwerkt zullen worden.

Als u nog opmerkingen en/of suggesties hebt ten aanzien van het onderzoek of deze vragenlijst, dan kunt u de rest van deze bladzijde hiervoor gebruiken.

Dankwoord
Curriculum Vitae
List of Publications

Bregje E. van Sleuwen

Dankwoord

Het is een eer om te promoveren op een onderzoek waaraan zoveel mensen hebben meegewerkt. Tegelijkertijd is het onmogelijk om al deze mensen persoonlijk te bedanken, wat niet afdoet aan hun bijdrage aan de totstandkoming van dit onderzoek.

Een speciaal woord van dank wil ik allereerst uitspreken aan alle ouders en kinderen die hebben deelgenomen aan het onderzoek. Zonder jullie vertrouwen in onze begeleiding en jullie bereidwilligheid ons onderzoeksteam drie keer te bezoeken had dit onderzoek niet plaats kunnen vinden. Het ga jullie goed!

Mijn promotoren, Prof.dr. Wietse Kuis en Prof.dr. Tom Schulpen.
Beste Wietse, dank voor je immer snelle, kritische maar altijd opbouwende commentaar op mijn manuscripten. In onze gesprekken gaf je steeds blijk van een positieve betrokkenheid bij ons onderzoek en bleef je vertrouwen houden in een goede afronding van dit proefschrift.
Beste Tom, je was mede door je immense ervaring in onderzoek naar culturele verschillen in verzorgingsgewoonten één van de initiatoren van dit onderzoek. Je bleef enorm betrokken, ook toen je zelf wat meer uit beeld was door verandering van werkplek. Zeker in de laatste fase heb ik me enorm gesteund gevoeld door je volledigheid van je correcties en je sturende rol en warme betrokkenheid bij de afronding van mijn proefschrift.
Mijn co-promotoren, dr. Monique L'Hoir en dr. Adèle Engelberts.
Beste Monique, jouw immer inspirerende, enthousiasmerende en positieve aanpak bij alles wat je doet zie je bij weinig mensen. Al vanaf het begin van onze samenwerking, toen ik stage bij je liep, werd ik daarmee 'besmet'! Nog 'even' een aanvraag indienen, een artikeltje schrijven of een presentatie maken was niet ongewoon, het werden dus regelmatig (gezellige) late avonden. Dirk is ook vast blij dat dit ei gelegd is! Het onderzoek is ontstaan uit jouw eigen nieuwsgierigheid naar het vinden van een wetenschappelijk goed onderbouwde en eenduidige interventie bij de begeleiding van ouders van excessief huilende zuigelingen, iets wat in Nederland nog niet voorhanden was. Ik kijk met veel genoegen terug op de afgelopen jaren van het onderzoek, de vele scholingen die we samen verzorgd hebben en de internationale congressen die we bezochten. Ik verheug me op een langdurige en minstens zo positieve samenwerking als we tot nogtoe gehad hebben!
Beste Adèle, gedurende de gehele looptijd van het onderzoek begeleidde je mij in het wetenschappelijk benaderen van de materie. Jouw nuchtere kijk op zaken relativeerde vaak mijn praktische benadering en door jouw ervaring en begeleiding bij het schrijven van wetenschappelijke artikelen heb ik veel geleerd. Bij het corrigeren van mijn manuscripten gaf je altijd bescheiden maar duidelijk aan wat er niet correct of wetenschappelijk genoeg aan was en ook zaken die ik vanuit de praktijk logisch vond nam jij niet zomaar aan als waarheid. En altijd had je gelijk! Met veel plezier hebben Monique en ik genoten van alle hartelijkheid van jou en Bert

in het prachtige Bunde, waar we, onder het genot van allerlei heerlijks en een goed glas wijn, met regelmaat een flinke slag gemaakt hebben! De herinnering aan de tocht achterin een Smart (inderdaad, die zijn eigenlijk geschikt voor 2 personen....) in de heuvels rondom Florence (Italië) op weg naar een wijnhuis met de meest heerlijke wijnen en olijfolie zal ik niet snel vergeten! Ook met jou hoop ik nog op vele jaren voortzetting van deze plezierige samenwerking!

I would like to express my gratitude to all members of the Assessment Committee, Prof.dr. Linda de Vries, Prof.dr. Paul Helders, Prof.dr. Gerben Sinnema, Prof.dr. Menno Reijneveld, Prof.dr. Pim Assendelft, and Prof.dr. Ian St. James-Roberts. Thank you for your thorough and fast assessment of my thesis and all your constructive annotations.

De begeleidingscommissie van het onderzoek, bestaande uit Dr. Richard van Lingen, Dr. Frank Brus, Prof.dr. Tom Schulpen, Prof.dr. Wietse Kuis, Dr. Adèle Engelberts en Dr. Monique L'Hoir, wil ik graag hartelijk danken voor hun waardevolle bijdrage bij de opzet en uitwerking van het onderzoek. Jullie hebben voorafgaand aan de start van het onderzoek een boel 'angels' weten te verwijderen, waardoor dit onderzoek een krachtig en wetenschappelijk sterk geheel is geworden.

De medewerkers van het onderzoek verdienen een *belangrijke* plek: Ria Blom, Kiki Mulder, Arianne Bronsvoort, Simonne Goderis, Gonnie Hoenkamp en Marlene Venderink. Dankzij jullie accuratie en 'denken vanuit de praktijk' in de aanloop naar het onderzoek is er een werkbaar programma ontstaan. Door jullie niet aflatende enthousiasme en overtuiging van het effect van beide interventies hebben ruim 400 ouders en kinderen zich in de periode van het onderzoek enorm gesteund gevoeld. Na afronding van het onderzoek hebben jullie met een evenveel verve samen met Monique en mij het overgrote deel van de medewerkers in de jeugdgezondheidszorg geschoold over de praktijktoepassing van regelmaat en de richtlijnen rondom inbakeren. We hebben dit samen voor elkaar gekregen!

De medewerkers van de jeugdgezondheidszorg, huisartsen, kinderartsen, (kinder-) fysiotherapeuten en alle overigen die excessief huilende zuigelingen verwezen hebben naar ons onderzoek en daarmee een deel van hun zorg voor deze kinderen uit handen hebben willen geven, verdienen een groot woord van dank. Zonder deze *inzet hadden* wij nooit in zo'n korte periode ruim 400 kinderen kunnen includeren!

De beide statistici Paul Westers en Wim Busschers hebben mij met regelmaat aan hun bureau getroffen. Door de enorme grote dataset was het zelfs nodig een aangepast programma te gebruiken voor de analyses. Dank dat jullie beiden altijd de tijd namen om de soms voor mij ingewikkelde statistische technieken begrijpelijk uit te leggen!

Gedurende het onderzoek hebben meerdere stagiaires een bijdrage geleverd aan het onderzoek. Anneloes van Bodegraven en Karin Postelmans hebben een pretest van de brochure 'Inbakeren brengt rust' uitgevoerd. Maaike Beltman gaf in een scriptie een literatuuroverzicht over het inbakeren van kinderen en analyseerde enquêtes ingevuld door ouders die hun kind ingebakerd hadden. Nienke Blom en Pauline van den Berg onderzochten de potentiële risicofactoren van ouders en het gezin op het ontstaan van excessief huilen. Louise Berkhout voerde mede de follow-up studie uit naar de groep kinderen die gedurende de eerste week van het onderzoek, de baseline week, positief reageerden op hypoallergene voeding (Hoofdstuk 5). Françoise de Bruin heeft een belangrijke bijdrage geleverd aan het follow-up onderzoek bij (voormalig) excessief huilende zuigelingen op de leeftijd van 3 - 5 jaar. Dank jullie allemaal, jullie hebben een belangrijke bijdrage geleverd aan dit onderzoek!
Bij het invoeren van een enorme berg gegevens uit de vragenlijsten en dagboeken heb ik veel hulp gekregen van Irene van de Laar, Anne Maayke Spreeuwers en mijn zus Maartje van Sleuwen. Dank voor jullie tomeloze inzet en tijd die jullie gestoken hebben in dit onderzoek!

Mede dankzij mijn collega's op de Afdeling Medische Psychologie van het Wilhelmina Kinderziekenhuis heb ik jarenlang met veel plezier aan mijn onderzoek gewerkt. Als onderzoeker op een afdeling die met name gericht is op patiëntenzorg voelde ik mij desondanks nooit een vreemde eend in de bijt en was eenieder van jullie steeds geïnteresseerd (vaak ook privé) in de materie en voortgang van het onderzoek. Jullie hebben tijdens de inclusieperiode vaak 'last' van mij gehad wanneer er op maandag ouders met een ontroostbare baby op de gang zaten te wachten. Ik wens jullie allen veel succes!
Voor mijn collega-onderzoekers en kamergenoten een apart woord van dank. Sascha Duijff, Petra Klaassen en Maaike Beltman, vaak hebben we gesproken over de uitvoering van onze onderzoeken maar minstens zo vaak kletsten we het weekend even door of bespraken we de nieuwe kunsten van onze kroost. Ik wens jullie een goede afronding van jullie eigen onderzoek toe met als kroon op jullie harde werken een prachtig proefschrift!
Onze secretaresse, Marlies, Evelien, Margreet, Mariëlle, Annemiek en Gabriëlle, hebben in belangrijke mate bijgedragen aan het soepel laten verlopen van de stroom ouders die zich aanmeldde voor het onderzoek, de drukbezochte maandagen met ouders en baby's op de gang en een altijd gezellig praatje bij binnenkomst. Dames, bedankt!

Met mijn huidige collega's bij TNO Kwaliteit van Leven gaat de sfeer die ik zo waardeerde in het WKZ gelukkig niet verloren! De belangstelling voor mijn onderzoek en promotie is hartverwarmend, de nieuwe ideeën en lopende onderzoeken zijn inspirerend. Ik ben er dan ook vast van overtuigd dat ik bij TNO volledig op mijn plek zit!

Annemarie Engelhart, jij ontwierp voor mijn proefschrift de prachtige kaft. De wensvlaggen staan voor regelmaat, voor de wens van ouders dat het huilen snel verdwijnt, voor inbakerdoeken (of wasgoed?). Dank je voor je mooie ontwerp en je creativiteit!

Dit onderzoek was niet mogelijk geweest zonder financiële ondersteuning. In dit kader wil ik allereerst graag ZonMw bedanken voor het in ons gestelde vertrouwen. Daarnaast bedank ik de Weijers Stichting, de Ridderlijke Duitsche Orde, Balije van Utrecht, de Stichting Onderzoek en Preventie Zuigelingensterfte (bij afkorting Stichting Wiegedood), het K.F. Heinfonds en het Wilhelmina Kinderziekenhuis.
Graag wil ik de bedrijven bedanken die een bijdrage hebben geleverd aan dit onderzoek: Friso Kindervoeding voor het beschikbaar stellen van de hypoallergene voeding (Frisolac Allergy Care), Weleda Nederland NV voor hun bijdrage aan de inbakerdoeken die gebruikt werden tijdens het onderzoek en Stichting Kinderpostzegels Nederland voor het financieren van de boekjes 'Regelmaat brengt rust' en 'Inbakeren brengt rust' die uitgedeeld werden tijdens de scholingen aan medewerkers in de jeugdgezondheidszorg na afloop van het onderzoek.

Lieve familie, vrienden en buren. Bedankt voor jullie interesse in de voortgang van het onderzoek en de hartelijke complimenten als er weer een mijlpaal bereikt was. Ik heb veel aan jullie gehad!

Mijn paranimfen Judith Veen en Maartje van Sleuwen waren beiden mijn getuigen bij mijn huwelijk met Wilbert. Ik ben er dan ook erg trots op dat jullie op deze belangrijke dag ook weer naast mij zullen staan. Lieve Judith, lieve Maartje, het wordt een traditie: samen op naar de volgende mijlpaal?

Lieve pap en mam, dankzij jullie heb ik de kans gehad te studeren. Jullie waren er altijd een groot voorstander van om eruit te halen wat er in je zit. Door tijdens mijn onderzoeksaanstelling op het juiste moment een prikkelende opmerking te plaatsen ('maar als je er geen zin meer in hebt, dan stop je er toch gewoon mee?') gaven jullie mij toch net dat ene zetje om, koppig als ik ben, te laten zien dat ik het juist wél af zou ronden. Ook de dagen dat jullie met onze meiden op stap gingen zodat ik kon schrijven waardeer ik erg. Dank jullie wel, ik ben er trots op dat ik jullie dochter ben!

Lieve, lieve Wilbert. Als laatste in de rij, maar voor mij op de eerste plaats. Tijdens alle jaren van het onderzoek, maar met name in de laatste periode, heb jij mij vaak ontlast door op zaterdag- en zondagochtend op stap te gaan met Juul en Guusje zodat ik kon werken. Je baalde dat ik niet met jullie mee kon, samen is veel leuker, terwijl je toch enorm genoot van je kleine meiden. Wat hebben we straks lekker veel tijd over, tijd voor elkaar, voor de meiden: het lijkt me heerlijk! Hoewel, met nog twee kleintjes erbij in oktober zal deze rust van korte duur zijn..... Laten we dan nu samen genieten van deze grote overwinning. Alle hulde komt jou toe!

Curriculum Vitae

Bregje Elisabeth van Sleuwen werd geboren op 8 maart 1977 te Heeswijk-Dinther. Zij deed in 1995 eindexamen Gymnasium Bèta aan het Gymnasium Bernrode te Heeswijk-Dinther. In september 1995 startte zij haar studie Biologie aan de Universiteit Utrecht. In 1999 liep zij in het kader van haar specialisatie GezondheidsVoorlichting en -Opvoeding (GVO) stage in het Wilhelmina Kinderziekenhuis (WKZ) en verrichtte zij onder leiding van Dr. M.P. L'Hoir een onderzoek naar 'De implementatie en het gebruik van de folder Veilig Slapen (Stiching Wiegedood) op het consultatiebureau'. Hierna volgde een onderzoek naar 'Verzorgingsgewoonten in relatie tot wiegendood in Turkse, Marokkaanse en Nederlandse gezinnen in Nederland', waarin zij begeleid werd door Prof.dr. T.W.J. Schulpen, Dr. M.P. L'Hoir, en Dr. A.C. Engelberts. Gelijktijdig met dit onderzoek liep zij stage op de Afdeling Kinderpsychiatrie in het Academisch Ziekenhuis Utrecht (het huidige UMC Utrecht). In augustus 2000 behaalde zij haar Doctoraal Examen Biologie en aansluitend startte zij haar promotieonderzoek naar 'De effectiviteit van inbakeren bij baby's die excessief huilen; een gerandomiseerd, gecontroleerd onderzoek' op de Afdeling Medische Psychologie & Maatschappelijk Werk van het WKZ. In oktober 2005 startte zij samen met Dr. L'Hoir een landelijk scholingsprogramma over 'Regelmaat en inbakeren' voor medewerkers in de jeugdgezondheidszorg (artsen en verpleegkundigen consultatiebureau), waarvan inmiddels meer dan 50 van de 61 jeugdgezondheidszorginstellingen de scholing gevolgd heeft. Daarnaast maakte zij deel uit van de Werkgroep Inbakeren (ActiZ, organisatie van zorgondernemers) waarmee zij de concept-nota 'Aanpak van excessief huilen bij zuigelingen', gebaseerd op de resultaten uit haar promotieonderzoek, opstelde. In januari 2006 werd zij benoemd tot secretaris van de ZonMw Werkgemeenschap Jeugd & Gezondheid. In maart 2006 trad zij toe toe het bestuur van de Stichting Onderzoek en Preventie Zuigelingensterfte, bij afkorting Stichting Wiegedood. In juni 2006 werd zij samen met Dr. M.P. L'Hoir, Dr. A.C. Engelberts en Drs. I.M.B. Russel genomineerd voor de Jan Brouwersprijs 2006 tijdens het congres 'Ouders helpen opvoeden'. In oktober 2006 introduceerde zij samen met Dr. L'Hoir het landelijke scholingsprogramma 'Regelmaat, voorspelbaarheid en prikkelreductie' voor verloskundigen en kraamverzorgenden en in september 2007 het landelijke scholingsprogramma 'Regelmaat en inbakeren' voor medewerkers van de kinderafdelingen in algemene en academische ziekenhuizen. In augustus 2007 trad zij in dienst bij de sector Jeugdgezondheidszorg 0-19 jaar van TNO Kwaliteit van Leven, Preventie en Zorg.

In 2005 trad zij in het huwelijk met Wilbert P.M. Raap en werd, in tegenstelling tot het onderzoek geheel volgens planning, 9 maanden later hun eerste dochter Juul Elisabeth geboren. Na ruim anderhalf jaar werd het gezin uitgebreid met de komst van Guusje Johanna. Eind 2008 verwachten zij een tweeling.
In haar spaarzame vrije tijd verbouwde zij samen met Wilbert hun 'dijkwoning aan de Maas uit 1897 met authentieke details', was zij achtereenvolgens secretaris en voorzitter van de activiteitencommissie en redactielid van het clubblad van een watersportvereniging, coördinator van het jeugdkamp regio Utrecht/Flevo en Noord Holland van de Nederlandse Coeliakie Vereniging voor kinderen van 8 tot 16 jaar met coeliakie, secretaris van een bewonersvereniging in haar woonplaats Lith en lid van de Oudercommissie van het kinderdagverblijf van Juul en Guusje.

List of publications

- Sleuwen BE van, L'Hoir MP, Engelberts AC, Westers P, Schulpen TWJ. Infant care practices related to cot death in Turkish and Moroccan families in the Netherlands. Arch Dis Child 2003;88:784-788.
- Sleuwen BE van, L'Hoir MP, Engelberts AC, Busscher WB, Westers P, Blom MA, Schulpen TWJ, Kuis W. Comparison of behavior modification with and without swaddling as interventions for excessive crying. J Pediatr 2006;149:512-517.
- Sleuwen BE van, Engelberts AC, Boere-Boonekamp MM, Kuis W, Schulpen TWJ, L'Hoir MP. Swaddling - A systematic review. Pediatrics 2007;120:e1097-1106.
- Sleuwen BE van, Engelberts AC, Boere-Boonekamp MM, L'Hoir MP. Behaviour modification and swaddling as interventions to improve sleep: A link with obesity? Arch Dis Child jan 3rd 2007 (electronic pages);
- Long T. Abstract of: Sleuwen BE van, L'Hoir MP, Engelberts AC, Busscher WB, Westers P, Blom MA, Schulpen TWJ, Kuis W. Comparison of behavior modification with and without swaddling as interventions for excessive crying. Evidence-Based Nursing 2007;10:42
- Sleuwen BE van, L'Hoir MP, Engelberts AC. Reply to: Karp H. Swaddling and excessive crying, Letter to the Editor. J Pediatr 2007;151:e2-3
- Blom MA, Sleuwen BE van, Vries H de, Engelberts AC, L'Hoir MP. Regularity and swaddling – interventions that are applied when infants cry excessively. *Submitted for publication.*
- Sleuwen BE van, Berkhout L, L'Hoir MP, Schulpen TWJ, Kuis W, Meijer Y, Engelberts AC. Characteristics of excessively crying infants who react positively to a hypoallergenic diet. *Submitted for publication.*
- Sleuwen BE van, Engelberts AC, Schulpen TWJ, Kuis W, L'Hoir MP. Psychological factors related to the development of infantile excessive crying. *Submitted for publication.*
- Sleuwen BE van, Engelberts AC, Schulpen TWJ, Kuis W, L'Hoir MP. Do mothers of excessively crying infants differ psychologically from mothers of moderately crying infants? *Submitted for publication.*

Chapters in books

- Sleuwen BE van, L'Hoir MP. Excessief huilende zuigelingen. In: Es JC van, Keeman JN, Leeuw PW de, Zitman FG (red.), Het Medisch Jaar 2002: 17-37.
- L'Hoir MP, Sleuwen BE van. Huilbaby's. In: Bakker I. (red.), Kind & Adolescent, problemen en risicosituaties. Bohn, Stafleu, van Loghum, 2003.

Abstracts

- Sleuwen BE van, L'Hoir MP, Engelberts AC. Swaddling, an effective intervention? Congress Infant Crying, Padova, Italië, 16 juni 2002
- Sleuwen BE van, L'Hoir MP, Engelberts AC, Busschers WB, Westers P, Schulpen TWJ, Kuis W. Swaddling as intervention for excessive crying. 7th SIDS International Conference, Florence, Italië, 31 augustus - 4 september 2002
- Sleuwen BE van, L'Hoir MP. Excessief huilende baby's. Symposium Onderzoek in de Jeugdgezondheidszorg, wie durft?, Nijmegen, 20 September 2002
- Sleuwen BE van. Inbakeren van huilbaby's. SSPK, Zoetermeer, 21 maart 2003

- Sleuwen BE van. Swaddling, an effective intervention? SIDS European Conference, Oslo, Noorwegen, 28 – 31 juni 2003
- Sleuwen BE van. Swaddling, an effective intervention? Congress Infant Crying. Turku, Finland, 17 juni 2004
- Sleuwen BE van, L'Hoir MP, Engelberts AC, Busscher WB, Westers P, Blom MA, Schulpen TWJ, Kuis W. Swaddling, an effective intervention? 8th SIDS International Conference, Edmonton, Canada, 2 – 6 juli 2004
- Semmekrot BA, Engelberts AC, Mulder JC, Bijlmer RPGM, Joosten KFM, Liem KD, Sleuwen BE van, L'Hoir MP. ALTE in the Netherlands. 8th SIDS International Conference, Edmonton, Canada, 2 – 6 juli 2004
- Beltman M, Russel I, L'Hoir MP, Rijpert M, Sleuwen BE van. Child abuse in the Netherlands. 8th SIDS International Conference, Edmonton, Canada, 2 – 6 juli 2004
- Sleuwen BE van, L'Hoir MP. Comparison of behaviour modification with and without excessive crying as interventions for excessive crying. Master class professor Virginia Moyer, EMMA Kinderziekenhuis, Amsterdam, 13 januari 2006
- Sleuwen BE van, L'Hoir MP, Engelberts AC, Busschers WB, Westers P, Schulpen TWJ, Kuis W. Comparison of behaviour modification with and without excessive crying as interventions for excessive crying. 24th Annual conference on sleep disorders in infancy and childhood, Palm Springs, USA, 25 – 27 januari 2006
- Sleuwen BE van, L'Hoir MP, Engelberts AC, Busscher WB, Westers P, Blom MA, Schulpen TWJ, Kuis W. Comparison of behavior modification with and without swaddling as interventions for excessive crying. 9th SIDS International Conference, Yokohama, Japan, 1 – 4 juli 2006
- L'Hoir MP, Engelberts AC, Sleuwen BE van, Schaijk M van, Houten AM van, Vlimmeren LA van, Carpenter B. Sleeping sacks and turning behavior of infants. 9th SIDS International Conference, Yokohama, Japan, 1 – 4 juli 2006
- L'Hoir MP, Sleuwen BE van, Russel I, Engelberts AC. Preventie van huilen voorkomt mogelijk schudden en slaan van een zuigeling. Congres Ouders helpen opvoeden, nominatie Jan Brouwersprijs 2006, 16 juni 2006
- Sleuwen BE van, L'Hoir MP, Bruin F de, Londen WM van. Is excessief huilen een voorbode voor probleemgedrag? Congres Is dit normaal, 26 januari 2007
- Sleuwen BE van, L'Hoir MP, Engelberts AC, Busschers WB, Westers P, Schulpen TWJ, Kuis W. Comparison of behaviour modification with and without excessive crying as interventions for excessive crying. 25th Annual conference on sleep disorders in infancy and childhood, Palm Springs, USA, 25 – 27 januari 2007
- Sleuwen BE van, L'Hoir MP, Engelberts AC, Busschers WB, Westers P, Schulpen TWJ, Kuis W. Swaddling as intervention for excessive crying. Congress Infant Crying, Copenhagen, Denemarken, 6 juli 2007
- L'Hoir MP, Sleuwen BE van, Engelberts AC, Schulpen TWJ, Kuis W. Voordracht: Swaddling: a systematic review. Congress Infant Crying, Copenhagen, Denemarken, 6 juli 2007
- Sleuwen BE van, L'Hoir MP, Engelberts AC, Schulpen TWJ, Kuis W. Landelijk onderzoek naar het effect van inbakeren bij excessief huilende zuigelingen; een follow-up na 3-5 jaar. NVK Congres Veldhoven, 2 november 2007

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