# Keeping families close

*Partners in neonatal care and research*

van Veenendaal, N.R.

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Keeping families close –Partners in neonatal care and research

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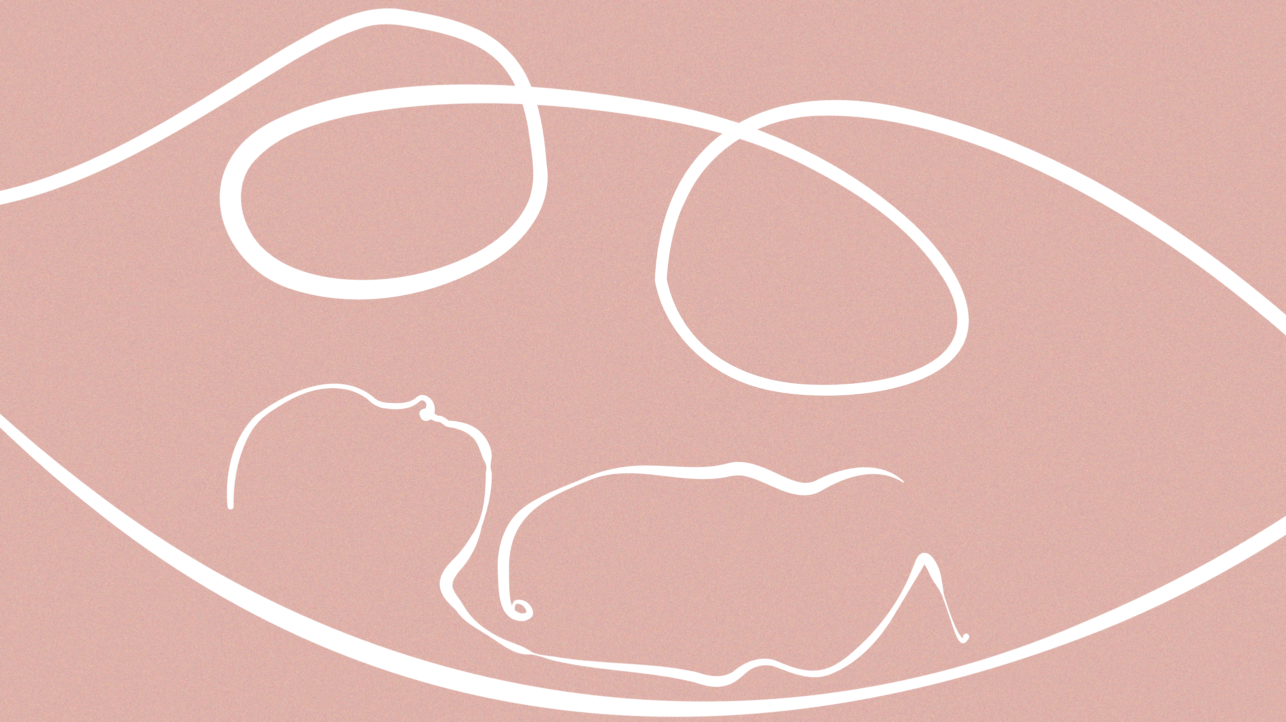


Part I

# Introduction - Facilitators and barriers to parent-infant closeness in the neonatal unit

Chapter 1

# Introduction

*Based on:*

**Effect of single family rooms for preterm infants on neurodevelopment: Study protocol for a systematic review**

*BMJ Open 2017;7:e015818*

**Nicole R. van Veenendaal,** Sophie R.D. van der Schoor, Jacqueline Limpens, Anne A.M.W. van Kempen, Johannes B. van Goudoever

&

**Family Integrated Care: supporting parents as primary caregivers in the Neonatal ICU**

*Pediatric Investigation 2021; 5 (2): 148-154*

Chandra Waddington, **Nicole R. van Veenendaal,** Karel O'Brien, Neil Patel on behalf of the International Steering Committee for Family Integrated Care,

## Preterm birth and the evolution of neonatal care

With the development of the incubator by professor Tarnier in the late 1800s, survival of preterm and ill infants increased enormously.[1] Shortly after, Professor Budin developed the first pavilion for "weaklings", the predecessor of the modern day neonatal intensive care unit (NICU).[2] Later, Incubator Baby Shows were of rage to display the progress in medical care, when chances were small for these vulnerable infants to survive.[1,3] Admission fees were asked to visitors, while mothers were exempt from paying these, but were not allowed to care for their infants while they were in the exhibit as caregiving was performed by highly skilled professionals.[4] With the evolution of neonatal care and improvement of life-saving technology over the past decades, the survival of ill and preterm (born before 37 weeks of gestation) infants has increased significantly and limits of viability have gone down.[5,6] Annually, approximately 15 million babies are born preterm, 11.1% of all livebirths worldwide. This percentage ranges from about 5-7% in several European countries, 12% in the USA, to 18% in some African and Asian countries.[7,8] Direct complications of preterm birth account for one million deaths each year, and preterm birth is a risk factor in over 50% of all neonatal deaths. Also, preterm birth and related morbidities result in high economic costs due to neonatal intensive care, follow-up care, special educational needs, and use of public sector resources.[9–12] In addition, preterm birth in itself can result in a range of long-term complications in survivors, with lifelong effects on metabolic, respiratory, neurological and physical health, which contribute to the prematurity-related burden of chronic disease in adults.[13,14]

In the modern environment of the NICU, preterm infants are exposed to multiple stressful experiences including painful diagnostic and therapeutic procedures (such as heel lances, breathing assistance, intravenous catheters) and disruptive environments (such as lighting or noise and separation from their parents).[15–18] Infants born preterm are particularly sensitive to stressful procedures because their neurologic system is immature and developing, and they cannot selectively limit or inhibit incoming stimuli and their physiological impact. This for instance includes their suboptimal reflexes and attention skills, state regulation difficulties, excitability, and hypo- or hyper-tonicity.[19–23] Negative stimuli affect the developing brain and its structure.[24] Accumulation of negative experiences in early life are associated with adverse neurodevelopmental and behavioural outcomes affecting language, motor function, and cognitive abilities into childhood on top of the burden of prematurity itself.[15,16,25–30]

**Parent-infant closeness and separation**

Parents can act as a buffer to stressful experiences in the NICU and they are vital and irreplaceable partners for the NICU team providing sensory cues to their infant (olfactory, auditory, visual and tactile) and aid in the regulation of pain, stress, warmth and development of their infant.[31–38] For instance, parents have a strong non-pharmacological role in pain management by providing skin-to-skin care, breastfeeding or "facilitated tucking".[39,40] As parents interact with their infant, this reciprocally stimulates the infant's neurobiological and behavioural processes (such as neurogenesis, neurite growth, and synaptic transmission), increases in oxytocin release, all supporting parent and infant well-being, parent-infant interaction in itself and the infant's response to pain and stress.[36,41–43] The early work of Bystrova et al. in a randomized trial of different care practices in Russia, compared "non-separation" with early skin-to-skin contact between mothers and their term infants and common separation practices of infants admitted to separate wards from their mothers for at least 2h hours after birth.[41,44,45] Non-separated mother-infant dyads had higher breastfeeding rates, better temperature regulation and improved mother-infant interaction 1 year later. Physical and emotional parent-infant closeness is therefore important, providing the neurobiological foundation for an infant's ability to form social bonds later in life and to ensure optimal physical, emotional, and mental wellbeing in the short and longer term in both infants as parents.[32,43,46–48]

However, hospitalization of preterm infants in the NICU, is often characterized by parent-infant separation.[31,49–52] Whilst and probably due to the rapid (technological) improvements in neonatal care over the past decades and the compartmentalization of newborn and maternity care, parental presence, participation and integration into neonatal care, and parent-infant closeness has still not always been fully implemented in the modern day NICU.[3,53–56] During hospitalisation of their infant in the NICU, in this highly technical environment with highly skilled staff, parents are usually assigned a visiting and supportive role. Parents have feelings of helplessness and powerlessness during this period, they cannot take on normal parenting roles and they experience trauma.[31,49–52,57–61] . Parental stress arising from the experience of the NICU is an important issue that potentially impacts parenting behaviour and long-term emotional and health problems in parents and their infants.[62–65] Contrarily, if parents are integrated as essential caregivers into neonatal care and parent-infant separation is minimized, parents can meet their infants' physical and developmental needs during hospitalization. Parents are then better prepared to take care of their infant at home, resulting in health benefits for both infants and their parents.[34,55,59,66–74]

Parent advocacy groups and the World Health Organization also widely and publicly strive for parental presence and integration of parents into neonatal care, as this is still not yet fully implemented.[63] As such, the European Standards of Care for Newborn Health and the World Health Organisation Survive and Thrive Report recommend to accommodate parents in skin-to-skin care (SSC), to actively welcome and engage parents in the care of their newborn, and to facilitate parental presence throughout the 24 hours by an optimal design of the NICU.[75–79] In addition, the European Foundation for the Care of Newborn Infants (EFCNI) states in their Rights of parents and newborns that "all families have the right to be considered as a unit [..], all parents have the right to receive appropriate education and be actively involved in their baby's care giving in an effective and sensitive manner" and that "all parents and newborns have the right to family-centered care and to stay together while the child receives healthcare" [76,77].

**Parent-partnered neonatal care, family centered care and family integrated care**

Within this thesis, we had an overall interest in parent-partnered neonatal care practices[70] and specifically family integrated care (FICare), which builds upon the principles of family centred care (FCC).[55] *Parent-partnered neonatal care models* are approaches that centre or integrate parents as full partners in the delivery of hospital care to their ill or small newborns as is explained in the taxonomy of Franck and O'Brien.[70] The models vary in emphasis, but all share common elements of specified parent-delivered interventions and roles and responsibilities of the clinical team and parents.[70] *Family centered care* has not been uniformly and strictly defined[70], but has at its core the principles of Respect and Dignity, Information Sharing, Family Support, Participation and Collaboration.[4,80–83] It entails a caregiving philosophy in pediatric and neonatal care with underlying core principles of mutual trust and power-sharing between parents and healthcare providers. It is grounded in the belief that partnerships among health care providers, patients, and families is mutually beneficial and leads to higher quality and safer healthcare.[80,81,84]

*Family integrated care* comprises a comprehensive framework to implement a family centred care philosophy by bringing parents, medical and nursing staff together to develop a collaborative program of education and support. It was adopted to the Canadian context by professor Shoo Lee and his team of Mount Sinai Hospital in Toronto from the "Truly Baby-Friendly Unit" with a care by parent unit in Estonia described by Levin et al. in the 1990s.[55,85–90] Levin et al. published on the benefits of this care-by-parent program, with 24 hour care by the mother, supporting breastfeeding and the use of human milk, and minimal use of technology and sparse contact between the baby and medical staff.[85] After, he also proposed a humane neonatal care initiative with 11 steps for the improvement of psychosocial and medical care in units for sick newborns, which can potentially be seen as the foundations for family centered and integrated care.[85,91,92] FICare has 4 pillars (Staff Education & Support, Parent Education, NICU Environment and Psychosocial Support) that endorses parent-provider partnership and parent-infant interaction by enabling parents as primary caregivers in the neonatal unit and as equal partners in the care and implementation team.[55,93–96] Within FICare, parents are perceived as part of the healthcare team, they receive educational sessions, they are included on daily rounds and in shared decision making, and they are asked to actively participate in the care for their infant.[70] Above, the NICU environment is supportive towards 24h parental presence and the healthcare staff is educated on the importance of parent involvement in care and special needs of these parents.[86,89] It shifts the paradigm of care so that neonatal staff provide education, mentorship, and coaching that focuses on the parent-infant dyad, where parents and infants are considered mutually dependent.

**Aims of this thesis**

This thesis discusses how we can ameliorate (adverse) outcomes (such as infections in infants and stress in parents) related to hospitalisation of preterm infants in the modern day NICU by adjusting and adopting the environment and family care giving practices for these vulnerable patients and their families. It discusses the setting and culture that is currently present in NICUs regarding family presence and participation in neonatal care.

**Part 1** of this thesis comprises the introduction into the different concepts of single family rooms (SFR), FICare, and parent-infant closeness. It also studies associated health outcomes in parents and infants and barriers and facilitators to successful implementation of parent-infant closeness and FICare. This is done by means of determining the effect of the environment, specifically of hospitalising preterm infants in single family rooms (SFR) versus open bay units (OBU) on primarily neurodevelopment with a systematic review and meta-analysis of the published literature **(Chapter 2)**. Secondly, we studied the association of FICare in SFR compared to standard care in OBU and late-onset sepsis in preterm infants in a retrospective cohort study with mediation analysis. Mediation analysis was applied, to understand pathways towards improved outcomes in the infants admitted to a setting with FICare in SFR. Peripheral- or central-venous catheters and parenteral nutrition were investigated as potential mediators of the association. Secondary outcomes included length of stay and exclusive breastfeeding at discharge **(Chapter 3)**. In a second systematic review and meta-analysis, we analyzed the effects of hospitalising preterm infants in single family rooms versus open bay units on parent outcomes, specifically NICU stress **(Chapter 4).** Little is known on the current state and application of mother- and parent-infant closeness during NICU-stay of the infant and which potential barriers health care professionals encounter to keep families close. Therefore, we sought to give an overview of current obstetric, maternity and neonatal settings with regard to parent-infant closeness and family integrated care practices during infant hospitalization in neonatal intensive care units in 18 European countries and Canada in a qualitative study with semi-structured interviews. We aimed to define themes for success for implementation of parent-infant closeness **(Chapter 5)**.

**Part II** includes the operationalization of participation, collaboration, and communication of parents with health care providers within the NICU context. This is done, by establishing a psychometrically validated tool to capture the concept of parent participation and collaboration in neonatal care (**Chapter 6).** In the NICU parents continually engage in communication with healthcare staff. Parents' experiences with communication during their infants' admission can contribute to relieving or increasing their distress. Communication can serve important clinical goals such as relaying information, obtaining consent, making decisions and foster collaboration between parents and staff. We therefore sought to give a systematic overview of the precise role and functions of communication introducing the NICU Communication Framework, to discuss what 'good' parent-provider communication precisely entails **(Chapter 7).** Following this framework, we aimed to methodically map the effects of parent-provider communication during infant hospitalization in the NICU on parent-related outcomes with a systematic review with meta-synthesis and narrative synthesis **(Chapter 8)**. We sought to contribute to theoretical conceptualizations of NICU communication and – ultimately – to improve the quality of parent-provider interaction and family integrated care implementation in practice.

**Part III** encompasses the clinical studies carried out in a prospective observational cohort study in level 2 neonatal wards in Amsterdam and Alkmaar, the Netherlands. In these studies, we studied the effect of an innovative FICare model and the association with outcomes in mothers and fathers of preterm infants. Within this facility, a multidimensional neonatal care model with complete couplet care for the mother-infant dyad, within single family rooms with additional FICare practices is implemented. We studied whether active participation and partnership in care were a potential mediator of the association between the FICare model and improved mental health outcomes in mothers of preterm infants (**Chapter 9).** Additionally, we sought to study the experiences of fathers in this facility. Many studies on FICare and experiences in the NICU focus solely on mothers of preterm infants. Fathers* -too- can feel stressed, depressed, excluded, isolated, and incompetent during and after hospitalisation of their infant in the NICU. Additionally, the effect of infant hospitalisation on fathers might be different than on mothers. Therefore, we studied the association of the innovative FICare model and mental health in fathers of preterm infants **(Chapter 10).**

This thesis concludes with a general discussion of the findings and future perspectives **(Chapter 11).** Finally, an English and Dutch summary is presented.



**We realize, recognize and respect that there are people having children who may not identify as father or mother. Therefore, we use the term fathers throughout this thesis for partners of the newborn's birthing mother who will assume a parental role and act as essential caregiver.*

# Chapter 2

# **Hospitalising preterm infants in single family rooms versus open bay units:** a systematic review and meta-analysis

*Lancet Child & Adolescent Health 2019;3:147–57.*

**Nicole R. van Veenendaal,** Wieke H. Heideman, Jacqueline Limpens, Johanna H. van der Lee, Johannes B. van Goudoever, Anne A.M.W. van Kempen, Sophie R.D. van der Schoor

# ABSTRACT

### Background

The effect of the hospital environment on health and specifically neurodevelopment in preterm infants remains under debate. We assessed outcomes of preterm infants hospitalised in single family rooms compared to common open bay units.

### Methods

For this systematic review and meta-analysis, we searched MEDLINE, EMBASE, PsycINFO, the Cochrane Central Register of Controlled Trials (CENTRAL), Web of Science, and Clinicaltrials.gov from inception to the 13$^{th}$ of August 2018 using controlled terms (i.e. MeSH-terms) and text words related to prematurity and NICU design. We included randomised and non-randomised studies. Methodological quality was assessed using The Cochrane Collaboration's Risk of Bias Tool for randomised controlled trials and the Cochrane Risk of Bias Tool for Non-Randomised Studies of Interventions. Summary estimates for meta-analysis were calculated using random effects models.

### Findings

We identified 487 records. Thirteen study populations (n= 4,793) were included. No difference in cognitive neurodevelopment was found on the Bayley Scales of Infant and Toddler Development-III at 18–24 months of corrected age (n= 680 patients, MD+1·04, 95%CI -3·45, 5·52, $I^2$= 42%, p= 0·65). The incidence of sepsis was lower (n= 4,165 and 108,305 hospitalisation days, RR= 0·63, 95%CI = 0·50–0·78, $I^2$= 0%, p<0·0001) and exclusive breastfeeding at discharge was higher in single family rooms (n= 484, RR= 1·31, 95%CI = 1·07–1·61, $I^2$= 0%, p= 0·01).

### Interpretation

Single family rooms should be considered to hospitalise preterm infants, as sepsis is reduced and exclusive breastfeeding is higher. No difference in long-term neurodevelopment was detected.

# INTRODUCTION

Every year, 14·9 million infants are born preterm worldwide.[8] After birth, preterm infants can spend a considerable period in the neonatal ward before discharge to home. Currently, most preterm infants are hospitalised communally in open bay units (OBUs), often near the maternity ward, but physically separated. Nurses provide routine care and parents are welcome in most units at all times. Due to the setting, the emotional and physical closeness between caregivers and their infant, and parental presence during infant stay is potentially limited.

Unfavorable environmental factors (such as separation from parents, lighting, noise, exposure to varying levels of pain, and pain-related stress) during hospitalisation and immaturity itself, might influence the range of morbidities in preterm infants, including neurodevelopment and psychosocial behavior.[26,28] Especially, infections during hospital stay jeopardise the survival and neurodevelopmental outcomes of these infants.[97] Preterm infants are prone to infection due to a decreased period of placental passage of maternal antibodies and due to their immature immune system.[98] Another challenge in the NICU includes the establishment of breastfeeding.[50] Breastfeeding has many beneficial effects on the preterm infant, but rates of exclusive breastfeeding are lower in preterm infants versus term infants at discharge from the hospital.[99]

We have noticed a movement in the neonatal field to build single family rooms (SFRs) for this vulnerable patient population, admitting one infant per room, enabling the parents to be present continuously, including during the night. The potential clinical benefits and harms of SFRs for the preterm infant are the subject of debate. A previous review studying preterm infants showed that interventions including parents (such as parent education, infant stimulation, home visits or individualised developmental care) enhanced neurodevelopment in this vulnerable patient group. However, this review did not include and assess the influence of SFRs.[94] A Cochrane review analysed the effect of rooming-in on breastfeeding rates, but this was limited to only healthy term infants.[100] The review by Shahheidari and colleagues discussed published research on SFRs between 2000 and 2011, without assessing outcomes in preterm infants specifically and without providing meta-analyses.[101] We asked parents in the Association of Veteran Parents of Infants admitted to the NICU in the Netherlands about the importance of SFR during NICU stay. Ninety-seven percent of respondents (n= 48) indicated that SFRs are important for the parents during NICU stay (unpublished data), adding to the rationale for this review.

In this study, we assess the difference in effect of hospitalisation in single family rooms versus open bay units in preterm infants, primarily on long-term neurodevelopmental outcome. Secondary outcomes were the length of stay, sepsis rates, growth, breastfeeding rates, mortality, retinopathy of prematurity (ROP), intraventricular hemorrhage (IVH), and bronchopulmonary dysplasia (BPD).

# METHODS

For this systematic review and meta-analysis, we used the Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA) guidelines.[102] This study was registered in PROSPERO (International Prospective Register of Systematic Reviews) on 2 November 2016 (registration number: CRD42016050643). A full protocol was published before conducting this research.[103] Deviations from the protocol are described in the Appendix.

**Search strategy, study selection, data collection and risk of bias**

A medical information specialist, experienced in systematic reviews, searched the following databases from inception to the 13th of August 2018: MEDLINE, EMBASE, PsycINFO (through the OVID interface), the Cochrane Central Register of Controlled Trials (CENTRAL), Web of Science and Clinicaltrials.gov. We used both controlled terms (i.e. MeSH-terms in MEDLINE) and free text terms related to prematurity (i.e. “preterm birth”, “preterm infant diseases”, “low birth weight”, etc.) and NICU design (i.e. “single family room”, “single crib room”, “rooming-in”, “open bay”, “hospital design and construction”, etc.). See Appendix for the full search strategies. There were no restrictions on the language, date, study type or publication status . We cross-checked reference lists and citing articles of identified relevant papers.

We included randomised clinical trials, cohort studies, quasi-experimental studies and before-and-after series. Two researchers (NvV and SvdS) independently screened abstracts and assessed full text articles for inclusion. We required studies to report a comparison between SFRs and OBUs and to provide summary data on clinical outcomes in preterm infants. We did not restrict our investigation to the primary outcome measure (neurodevelopment). Outcome data on parents (i.e. parental stress, satisfaction, depression etc.) were not the objective of this review. We calculated kappa and specific agreement for the screening of studies.[104] We collected data as described.[103] We contacted study authors twice for clarification of (missing) data in included and potentially eligible studies, specifically requesting data on preterm infants in mixed patient cohorts or missing data in abstracts only. We used the most complete and recent paper if multiple papers assessed the same (sub)population. Four reviewers applied The Cochrane Collaboration's Risk of Bias Tool for randomised controlled trials and the Risk of Bias in Non-randomised Studies of Interventions (ROBINS-I) tool to each study. These reviewers applied the risk of bias (RoB) tools twice; once on the outcome level (neurodevelopment) and once on the study level. This was done separately and independently.[105]



### Outcome measures

The primary outcome was the age-appropriate neurodevelopmental outcome of infants from nine months onwards measured by standardised scales for neurodevelopment.[94,103,106] Length of stay was defined as the length of hospitalisation from birth to discharge home. We recorded sepsis as defined by the authors. We calculated sepsis per total of patient days in the study (mean length of hospital stay in days × number of patients) and per 1,000 days of hospitalisation ((sepsis events/total patient days) × 1,000). Following Jaafar and colleagues, we classified breastfeeding as either *Exclusive breastfeeding* (an infant receiving only breast milk, without any additional formula feeding) or *Any breastfeeding* (an infant receiving any amount of breast milk regardless of the amount of additional formula feeding).[100] We calculated growth during hospital stay if data was available on birthweight, weight at discharge and length of stay. We defined mortality as death during hospital stay as reported or calculated from flow-charts. BPD was defined as the need for supplemental oxygen at 36 weeks of postmenstrual age or as described by the authors. IVH was a presence of at least grade III or IV.[107] ROP was described as defined by the authors.

### Statistical analysis

We used Review Manager (version 5.3; the Cochrane Collaboration) to conduct the meta-analysis . We assessed heterogeneity with the $I^2$ test for heterogeneity. We used a random-effects model for meta-analysis if heterogeneity was assessed to be acceptable. Continuous data were analysed by computing the weighted mean difference with 95% confidence intervals (CI). We calculated means and variances if this was not provided in the original publication as described previously.[108] For dichotomous outcomes; we calculated risk ratios (RR) with 95% confidence intervals.[109] We performed sensitivity analyses, and used meta-regression (with the "meta" and "metafor" package in R, version 3.4.3, Nov 2017) to estimate the effect of different assumptions on outcome variables (prespecified were RoB, gestational age at birth, and start of SFR care).[110] For outcomes with more than ten studies we assessed publication bias with funnel plots.

### Role of the funding source

The sponsor of the study had no role in study design, data collection, data analysis, data interpretation, or writing of the report. NvV, SvdS and AvK had full access to all the data in the study and had final responsibility for the decision to submit for publication.

## RESULTS

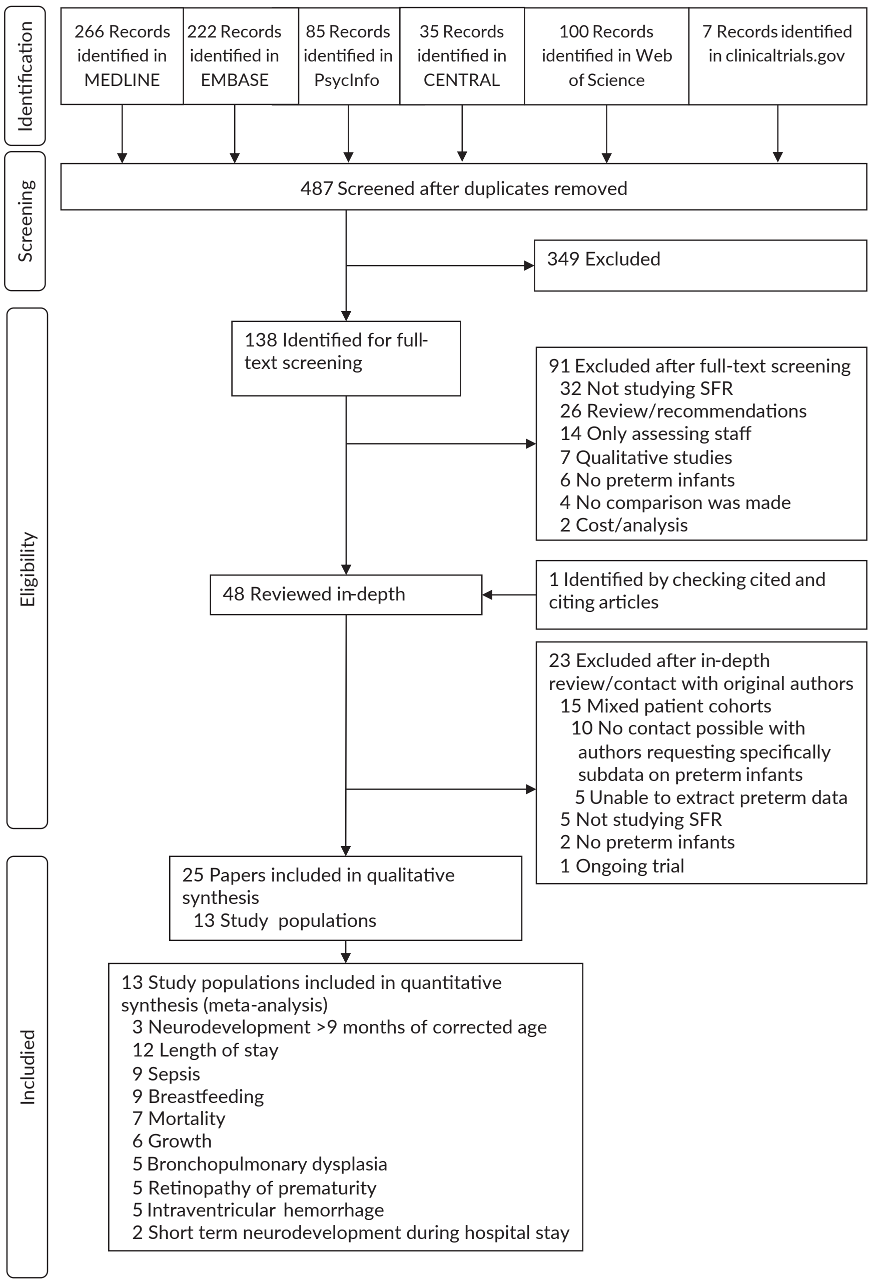
A total of 487 records were identified through the search. One-hundred-thirty-eight references were identified for full text screening (Figure 1). The inter-rater reliability for selection on titles and abstracts was good (Cohen's kappa: 0·87) and positive specific agreement (87·8%) and negative specific agreement (98·7%) for the screening of studies for eligibility was high. Ten studies needed additional discussion between the two reviewers and a third reviewer for in/exclusion. One additional paper was identified through cross-referencing. Forty-eight papers were reviewed in-depth. We contacted authors for additional information; 25/35 (71%) of the authors responded.

In total, 13 distinct study populations (total patient n= 4,793) were discussed in 25 papers (including five abstracts, Table 1). Seven study populations were described in multiple papers (Table 1 and Appendix). We identified one randomised trial[111,112], four non-randomised prospective studies[113–119], one non-randomised retrospective study[120], and seven populations before- and after relocation to a new hospital environment.[121–135] We used the propensity matched outcomes for meta-analysis for the cohort of Stevens and colleagues as this was described to be the best controlled outcome data (not available for ROP).[128] One paper was written in French,[120] all other papers in English. The papers were published between 2004 and 2018 and performed in developed countries.

### Risk of bias

All studies on the primary outcome had a moderate risk of bias (RoB, Table 2). Specifically, bias due to confounding and selection bias was of importance. Bias due to confounding was low in randomised trials and before-after studies within the same hospital.[111,112,121,125,126,128,130–132] Studies that described non-randomised trials within the same hospital were considered at moderate risk[115,116,119,120], and studies that described non-randomised trials between different hospitals were considered at serious risk.[113,114,117,118] Selection bias was considered serious if parents were asked to participate and to consent to the study and if this was dependent on their presence in the NICU.[113,114,117,118] Parental presence (and thus consent) could be influenced by the intervention studied (parents could possibly be present more in the SFR design). One study was considered at low RoB in all domains, a randomised controlled trial.[111,112] All other studies were of moderate to serious RoB. Three studies (including two abstracts) had insufficient information on at least one domain.[121,125,132]

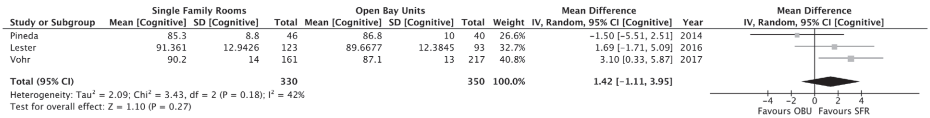
**Figure 1. Flow diagram**



## Single family rooms and the effect on neurodevelopment

Five different papers assessed the influence of SFR on short and long-term neurodevelopment (see Table 1).[115,119,124,134,135] Three papers included moderate long-term follow-up data using the BSID-III for evaluation of neurodevelopment at the corrected age of 18–24 months, all had >70% follow-up rates (Appendix). [115,124,135] The papers by Lester (2016) and Vohr (2017) partially reported on the same cohort (Appendix). The paper by Monson (2018) described some infants of the cohort of Pineda and colleagues.[115,116,119] All populations were extremely preterm (mean gestational age <28 weeks) with extremely low mean birthweights (<1000 grams). The effect of SFR on the cognitive composite score was not significantly different (Table 3 and Figure 2). Statistical heterogeneity was substantial (77% and 87%) for the motor and language composite scores, and therefore no meta-analysis was done for these subscales (Appendix).[115,124,135] A sensitivity analysis, including only Vohr (2017) and omitting Lester (2016) due to partial overlap, did not change this outcome and resulted in higher heterogeneity (Table 3).

**Figure 2. Single family rooms versus open bay units and the effect on longterm cognitive neurodevelopment**




SFR: single family room, OBU: open bay unit

## Secondary outcomes

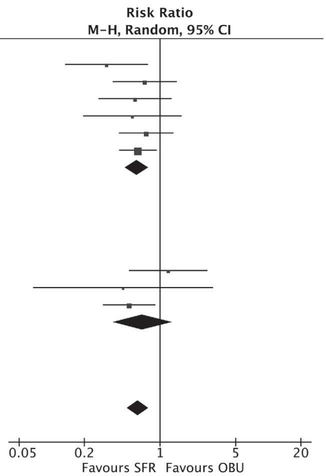
The mean length of stay varied between 10·2 and 84·4 days in twelve populations. We did not find a statistically significant difference in length of stay (n=4,702 infants, MD -1·33, 95%CI = -2·77, 0·11, p = 0·07, $I^2$ = 75%, Table 3).[111,113–115,118,120,121,124,125,128,130–132] Including only studies with low or moderate RoB did not alter this outcome (MD -2·46, 95%CI = -5·89, 0·97, p = 0·16, $I^2$ = 82%).[111,115,120,124,128,130] If only studies with low RoB due to confounding and due to selection of participants were included, heterogeneity improved, but no difference in length of stay was observed (n=3,495, MD -0·41, 95%CI = -4·67, 3·85, p = 0·85 ,$I^2$= 66%, Appendix).[111,124,128] Mean gestational age at birth explained 20% of heterogeneity in length of stay in meta-regression analysis. Although not statistically significant, for each additional week of gestational age at birth the difference in length of stay decreased with 0·2652 days (95%CI = -0·0297, 0·560, p = 0·078). Start of single room care (at admission or when the infant was stable) did not explain the heterogeneity in length of stay. Overall, lower sepsis rates were present in SFR (n= 4,165, RR = 0·63, 95%CI = 0·50–0·78, p <0·0001, $I^2$ = 0%,

Figure 3).[111,115,117,120,121,124,125,128,130] Analysing 108,106 patient hospitalisation days, sepsis rates were 2·92/1,000 hospital days in OBU and 1·95/1,000 in SFR. Including only studies with culture proven sepsis, this effect was persistent (n=3,968, RR = 0·62, 95%CI = 0·48–0·80, p = 0·0002, Table 3, Appendix).[111,115,121,124,128,130] Assessing only studies with overall low or moderate RoB, this effect was still present (n= 3,859, RR = 0·65, 95%CI = 0·50–0·85, p = 0·001, $I^2$ = 0%, Table 3)[111,115,120,124,128,130]. Also if only studies with low RoB due to confounding and selection bias were included (Appendix). We found a significantly higher incidence of exclusive breastfeeding at discharge in SFRs (n= 484, RR = 1·31, 95%CI = 1·07–1·61, p = 0·01, $I^2$ = 0%, Table 3 and Appendix).[114,117,120,131,132] One study was at overall moderate RoB[120] , and no studies were available with low RoB due to confounding and selection bias for this outcome (Appendix).



**Figure 3. Single family rooms versus open bay units and the effect on overall sepsis incidence during hospitalization**

| Study or Subgroup | SFR Events | SFR Total | OBU Events | OBU Total | Weight | Risk Ratio M–H, Random, 95% CI | Year |
|---|---|---|---|---|---|---|---|
| **Culture proven sepsis** | | | | | | | |
| Rosenblum | 6 | 106 | 18 | 102 | 6.4% | 0.32 [0.13, 0.78] | 2004 |
| Ortenstrand | 13 | 183 | 18 | 183 | 10.7% | 0.72 [0.36, 1.43] | 2010 |
| Stevens | 10 | 1300 | 17 | 1300 | 8.3% | 0.59 [0.27, 1.28] | 2011 |
| Domanico | 5 | 81 | 9 | 81 | 4.5% | 0.56 [0.19, 1.59] | 2011 |
| Pineda | 15 | 58 | 17 | 49 | 14.8% | 0.75 [0.42, 1.33] | 2014 |
| Vohr | 28 | 205 | 70 | 320 | 30.9% | 0.62 [0.42, 0.93] | 2017 |
| **Subtotal (95% CI)** | | **1933** | | **2035** | **75.5%** | **0.62 [0.48, 0.80]** | |
| Total events | 77 | | 149 | | | | |
| Heterogeneity: $Tau^2 = 0.00$; $Chi^2 = 2.80$, df = 5 (P = 0.73); $I^2 = 0\%$ | | | | | | | |
| Test for overall effect: Z = 3.70 (P = 0.0002) | | | | | | | |
| **Use of antibiotics** | | | | | | | |
| Wataker | 10 | 36 | 7 | 30 | 7.1% | 1.19 [0.52, 2.75] | 2012 |
| Chabaud | 2 | 68 | 2 | 31 | 1.4% | 0.46 [0.07, 3.09] | 2012 |
| Hourigan | 8 | 18 | 12 | 14 | 16.0% | 0.52 [0.30, 0.91] | 2018 |
| **Subtotal (95% CI)** | | **122** | | **75** | **24.5%** | **0.69 [0.37, 1.29]** | |
| Total events | 20 | | 21 | | | | |
| Heterogeneity: $Tau^2 = 0.11$; $Chi^2 = 2.97$, df = 2 (P = 0.23); $I^2 = 33\%$ | | | | | | | |
| Test for overall effect: Z = 1.17 (P = 0.24) | | | | | | | |
| **Total (95% CI)** | | **2055** | | **2110** | **100.0%** | **0.63 [0.50, 0.78]** | |
| Total events | 97 | | 170 | | | | |
| Heterogeneity: $Tau^2 = 0.00$; $Chi^2 = 5.61$, df = 8 (P = 0.69); $I^2 = 0\%$ | | | | | | | |
| Test for overall effect: Z = 4.12 (P < 0.0001) | | | | | | | |
| Test for subgroup differences: $Chi^2 = 0.11$, df = 1 (P = 0.75), $I^2 = 0\%$ | | | | | | | |




SFR: single family room, OBU: open bay unit

Higher rates of any breastfeeding at discharge were found (n= 1,109, RR = 1·06, 95%CI = 1·00–1·13, p = 0·04, $I^2$ = 0%, Appendix)[112,114,116,117,124,130,131], but this outcome became non-significant after sensitivity analyses for RoB (Table 3 and Appendix). No difference was found for mortality, BPD and IVH grade III-IV (Table 3).[111,114,115,121,124,128,130] We found a non-statistically significant lower incidence of ROP in children cared for in SFRs.[111,115,121,124,128] We did not find evidence for publication bias for length of stay (Appendix). The number of studies for other outcomes was insufficient for a definite conclusion on publication bias. In six studies, growth during hospital stay was between 15–23 g/day for infants in SFR versus 12–22g/day for children in OBU.[114,121,123,124,130,132] We did not perform meta-analysis as SDs were unavailable, and studies reported growth differently. The short-term neurological outcome was assessed in five papers of 2 distinct populations.[115,119,124,134,135] At 34 weeks of postconceptional age, Pineda and colleagues found higher levels of arousal on The NICU Network Neurobehavioral Scale (NNNS) in infants in SFR compared to OBU, also after controlling confounders .[115] Monson and colleagues described no difference in gray or white matter diffusion rates on MRIs.[115,119] At discharge, Lester and colleagues demonstrated that, subscores on attention were significantly higher and scores on physiologic stress, hypertonicity and lethargy were significantly lower in the SFR as opposed to the OBU.[134] Increased developmental support in SFR was a mediator for better attention and maternal involvement was a mediator for improved short- and longterm neurodevelopment.[134,135] Parental presence and participation was described in six populations.[111,113–116,118,131,134,135] It was measured differently between studies (Appendix) and no meta-analysis was performed. One study found lower parental presence in SFR if the mother was not hospitalised[113,114] and another found decreased time of skin-to-skin contact per day.[118] Overall, in four out of six populations, investigators noted an increase in parental presence, participation and skin-to-skin contact in SFRs compared to OBUs.[111,115,116,131,134,135]

## DISCUSSION

In this review and meta-analysis, we detected no difference in the long-term neurodevelopmental outcome in a small population of extremely preterm infants hospitalised in single family rooms or open bay units. However, we did find significantly less sepsis events during hospital stay, and higher rates of (exclusive) breastfeeding at discharge for infants cared for in single family rooms. This is of importance for all stakeholders in neonatal care.

All studies assessing neurodevelopment were at equal RoB and had homogenous study populations; the populations were extremely preterm (mean gestational age <28 weeks) with extremely low birth weights (<1000 grams). Outcomes in the individual studies showed that the mean BSID-III scores in this high-risk population were lower than the reference population (100), but comparisons between groups cared for in SFR versus OBU identified no differences. However, only two different distinct high-risk study populations with 464 individuals were assessed on this outcome. The mean difference was not only non-statistically significant, but the 95% CI did not exceed the effect size of 0.5, which is usually considered clinically relevant.

It might be hypothesized, that the preterm brain itself and factors related to prematurity -including antenatal, perinatal events and postnatal events like IVH and BPD- are the risk factors for a detrimental neurological outcome .[136] Therefore in this multifactorial context, a beneficial effect of SFRs alone is probably difficult to detect.[137]

Also, outcomes were not assessed after the corrected age of two years. As is stated by Wong et al. in an extensive meta-analysis of 3,133 children: "children who might experience cognitive difficulties at school-age were classified as having a normal neurodevelopmental function at ages one to three years. Even for cases of severe cognitive deficit, the accuracy in early detection was low".[138] More and longer follow-up studies are needed to assess the effect of SFRs in these children, as effects sometimes take a long time to develop. Our review shows that no follow-up studies have been conducted in infants >32 weeks of gestation, despite that these infants are also at risk for neurodevelopmental delay and morbidities.[139] Above all, late preterm infants make up of the majority (84·3%) of all preterm infants and add to the total burden of disease.[8]

Several studies tried to assess the factors that might be facilitated by implementing SFR, including parent participation and skin-to-skin contact. Parent participation in interventions and skin-to-skin contact has been shown to be beneficial in preterm and very low birth weight infants.[34,94] However, parent participation was not consistently recorded in the studies we analysed, due to the absence of a validated parent participation measurement tool, and it was only evaluated in one population as mediating factor for infant outcomes.[134,135]

Sepsis events were reduced with one per 1,000 hospital days in SFR in our analysis when compared to care in OBU. Also in sensitivity analyses considering culture proven sepsis and RoB in studies, the findings proved robust.

No studies described the association between the use of intravascular devices, skin-breaks and the incidence of sepsis in included studies.[97] This is an important issue for future research. Other possible explanations, such as antimicrobial use, intensity and frequency of patient contact by staff, skin-to-skin contact , and implementation of hygiene measures (specifically handwashing), should be investigated further.[34,140] Also overcrowding might be the explanatory factor, as a previous review in adult populations showed an association between overcrowding and hospital acquired infections.[141] A previous review focused on non-pharmacological preventive measures for infection in the NICU.[142] We add that the hospital environment should be considered in future studies.

We found a higher amount of breastfeeding at discharge for preterm infants hospitalised in SFRs. However, all studies were at moderate to serious RoB, and this outcome should be evaluated in methodologically more robust trials. Educational, socioeconomic, and lifestyle factors, such as smoking, are strongly associated with the mother's decision to breast-feed and also the WHO/UNICEF Baby Friendly Hospital Initiative has shown to promote breastfeeding.[143,144] In a previous Cochrane review only one RCT was included with low-quality evidence that rooming-in from birth until discharge from the hospital increased the rate of exclusive breastfeeding in *term* infants.[100] We add to this evidence that hospitalising preterm infants in SFR might increase breastfeeding rates at discharge. This might ameliorate health outcomes on the longer term for these vulnerable patients, but this needs further investigation.[143]

As the intervention studied is a change at the hospital unit level, many non-randomised studies were expected, and this was accounted for with the use of the ROBINS-tool, to elucidate potential sources of bias. With the inclusion of all different observational study designs, a complete overview has been created of the existing literature, with high generalisability for the neonatal field. However, only one randomised trial (with high internal validity) was found, and therefore no causal relationships could be given on the implementation of SFR on infant outcomes and this is a potential weakness of this review. This review had a pre-specified protocol, that was previously published and registered, adding to the transparency of this review conductance. We had an overall strong agreement on inclusions of studies into our systematic review with high kappa and specific agreements. Some papers assessed the same (sub)population, and as individual patient data was not extracted but summary estimates were used for this review, this is a potential limit of the review. We contacted the authors to provide us with additional information and 25/35 (71%) responded.

## CONCLUSION

This review and meta-analysis found no significant difference in long-term neurodevelopmental outcome between single family rooms and open bay units in a small population of extremely preterm infants. We found fewer sepsis events and more breastfeeding at discharge for preterm infants when hospitalised in single family rooms. Our findings support future developments towards building single family rooms and provides evidence for all players and stakeholders in the field of neonatal care.

# TABLES

**Table 1. Characteristics of studies and study-populations**

| Population | Source | Year | Country | Study design | Total in cohort, n | SFR, n | Start of care in SFR | OBU, n | Gestational age in SFR, mean (SD), wks, | Gestational age in OBU, mean (SD), wks, | Birth weight in SFR, mean (SD), grams | Birth weight in OBU, mean (SD), grams | *Assessed effect on:* *Neurodevelopment (longterm > 9mo)* | *Neurodevelopment (during hospital stay)* | *Length of hospital stay* | *Sepsis* | *Breastfeeding at discharge* | *Mortality* | *Growth* | *Parent participation/ presence* | *IVH* | *BPD* | *ROP* |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| 1 | [121] | 2004 | USA | BA | 208 | 106 | At admission | 102 | 29·4 (0·3) | 29·5 (0·3) | 1505 (52) | 1457 (48) | - | - | Y | Y | - | Y | Y | - | Y | Y | Y |
| 2 | [113,114] | 2008 | Turkey | NRPI | 60 | 31 | When stable | 29 | 30·8 (1·7) | 30·4 (2·1) | 1452 (82) | 1413 (351) | - | - | Y | - | Y | Y | Y | Y | - | - | - |
| | | 2009 | | | | | | | | | | | - | - | Y | - | - | Y | - | Y | - | - | - |
| 3 | [111] | 2010 | Sweden | RCT | 365* | 183 | When stable | 182 | 33·2 (2·8) | 32·8 (3·0) | 2097 (647) | 2021 (712) | - | - | Y | Y | - | Y | - | Y | Y | Y | Y |
| | [112] | 2012 | | | 289 | 152 | | 137 | 33·2 (2·8) | 32·6 (2·9) | NR | NR | - | - | - | - | Y | - | - | - | - | - | - |
| 4 | [130] | 2011 | USA | BA | 162 | 81 | At admission | 81 | 34·0 (NR) | 34·0 (NR) | 2282 (NR) | 2295 (NR) | - | - | Y | Y | Y | Y | Y | - | - | - | - |
| 5 | [128†] | 2011 | USA | BA | 2,600* | 1,300 | At admission | 1,300 | 35·24 (3·67) | 35·31 (3·67) | 2640 (910) | 2650 (880) | - | - | Y | Y | - | Y | - | - | Y | Y | Y |
| | [123] | 2011 | | | 73 | 42 | | 31 | 26·7 (1·3) | 28·1 (0·6) | 952 (44) | 1047 (47) | - | - | Y | - | - | - | Y | - | - | - | - |
| 6 | [116] | 2012 | USA | NRPI | 81 | 42 | At admission | 39 | 26·8 (0·8) | 26·3 (1·9) | NR | NR | - | - | Y | Y | Y | - | - | Y | Y | - | - |
| | [115] | 2014 | | | 107* | 58 | | 49 | 26·8 (1·6) | 26·4 (1·8) | 936·5 (263·8) | 953·6 (240·7) | Y | Y | Y | Y | - | Y | - | Y | Y | Y | Y |
| | [119] | 2018 | | | 46 | 24 | | 22 | 27 (2·1) | 26·3 (1·7) | 983 (250) | 883 (242) | Y | Y | Y | Y | - | - | - | - | - | Y | - |

**Table 1. Characteristics of studies and study-populations (continued)**

| Population | Source | Year | Country | Study design | Total in cohort, n | SFR, n | Start of care in SFR | OBU, n | Gestational age in SFR, mean (SD), wks, | Gestational age in OBU, mean (SD), wks, | Birth weight in SFR, mean (SD), grams | Birth weight in OBU, mean (SD), grams | *Assessed effect on:* *Neurodevelopment (longterm > 9mo)* | *Neurodevelopment (during hospital stay)* | *Length of hospital stay* | *Sepsis* | *Breastfeeding at discharge* | *Mortality* | *Growth* | *Parent participation/ presence* | *IVH* | *BPD* | *ROP* |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| 7 | 120 | 2012 | France | NRRI | 99 | 68 | At admission | 31 | 34·3 (0·3) | 34·1 (0·3) | 2182 (260) | 1892 (306) | - | - | Y | Y | Y | - | - | - | - | - | - |
| 8 | 117 | 2012 | Norway | NRPI | 66 | 36 | When stable | 30 | 32·7 (4·0) | 34·7 (2·2) | 1900 (600) | 2600 (838) | - | - | - | Y | Y | - | - | - | - | - | - |
| 9 | 118 | 2013 | Sweden | NRPI | 300 | 114 | When stable | 186 | 32·7 (2·9) | 32·1 (2·8) | 2046 (712) | 1980 (657) | - | - | Y | - | - | - | - | Y | - | - | - |
| 10 | 134 | 2014 | USA | BA | 403 | 252 | At admission | 151 | 28·3 (2·4) | 28·2 (2·3) | 1050 (278) | 1033 (261) | - | Y | Y | Y | - | - | Y | Y | Y | Y | Y |
| | 135 | 2016 | | | 216 | 123 | | 93 | 26·9 (1·7) | 27·1 (1·7) | 914 (220) | 938 (248) | Y | Y | Y | Y | - | - | Y | Y | Y | Y | Y |
| | 124 | 2017 | | | 530* | 210 | | 320 | NR; median: 27 | NR; median: 27 | NR, median: 980 | NR, median: 980 | Y | Y | Y | Y | Y | Y | Y | - | Y | Y | Y |
| 11 | 132 | 2015 | Australia | BA | 183 | 104 | At admission | 79 | 34·3 (range 28-36) | 34·5 (range 29-36) | 2238 (592) | 2321 (590) | - | - | Y | - | Y | - | Y | - | - | - | - |
| 12 | 131‡ | 2016 | Australia | BA | 81 | 32 | At admission | 49 | 32·9 (2·7) | 32·5 (5·3) | 1939 (691) | 1899 (495) | - | - | Y | - | Y | - | - | Y | - | - | - |
| 13 | 125§ | 2018 | USA | BA | 32 | 18 | At admission | 14 | 33·2 (2·2) | 31·4 (4·15) | 1949 (592) | 1704 (806) | - | - | Y | Y | - | - | - | - | - | - | - |
| *Total distinct patients measured* | | | | | 4,793 | 2,341 | | 2,452 | *Total distinct populations* | | | | 2 | 2 | 12 | 9 | 9 | 7 | 6 | 6 | 5 | 5 | 5 |

*:used for the population total and deemed the most complete report of the population (in papers describing the same patient populations; see Appendix)), †: includes[122,129], ‡: includes 2 abstracts[127,133], §: includes 1 abstract[126], BA: before-after study, BPD: bronchopulmonary dysplasia, IVH: Intraventricular Hemorrhage, n: number, NR: not reported, NRPI: Non-Randomised Prospective Intervention Study, NRRI: Non-Randomised Retrospective Intervention study, OBU: Open Bay Unit, RCT: Randomised Controlled Trial, ROP: Retinopathy of Prematurity, SD: Standard deviation, SFR: Single Family Room, wks: weeks, Y: Yes.

**Table 2. Risk of bias in included studies**

| Source | Bias due to confounding | Bias in selection of participants into the study | Bias in the classification of intervention | Bias due to deviations from intended interventions | Bias due to missing data | Bias in the measurement of outcomes | Bias in selection of reported result | The overall risk of bias |
|---|---|---|---|---|---|---|---|---|
| *Outcome level: Neurodevelopment (18-24 months of age)* | | | | | | | | |
| 115 | Moderate | Low | Low | Low | Moderate | Low | Moderate | Moderate |
| 135 | Low | Low | Low | Low | Moderate | Moderate | Moderate | Moderate |
| 124 | Low | Low | Low | Low | Moderate | Moderate | Moderate | Moderate |
| *Study level: for all secondary outcomes* | | | | | | | | |
| 121 | Low | No information | Low | Low | No information | Moderate | No information | No information |
| 113,114 | Serious | Serious | Low | Low | Low | Low | Moderate | Serious |
| 130 | Low | Moderate | Low | Low | Low | Low | Moderate | Moderate |
| 122,123,128,129 | Low | Low | Low | Low | Low | Low | Moderate | Moderate |
| 120 | Moderate | Low | Low | Low | Low | Low | Moderate | Moderate |
| 117 | Serious | Serious | Low | Low | Serious | Serious | Moderate | Serious |
| 115,116,119 | Moderate | Low | Low | Low | Low | Moderate | Moderate | Moderate |
| 118 | Serious | Serious | Low | Low | Low | Moderate | Moderate | Serious |
| 132 | Low | No information | Low | Low | No information | No information | No information | No information |
| 131 | Low | Serious | Low | Low | Serious | Serious | Moderate | Serious |
| 124,134,135 | Low | Low | Low | Low | Low | Moderate | Moderate | Moderate |
| 125,126 | Low | No information | Low | Low | No information | No information | Moderate | No information |
| **Cochrane** | **Bias arising from the randomization process** | **Bias due to deviations from intended interventions** | **Bias due to missing outcome data** | **Bias in the measurement of the outcome** | **Bias in selection of the reported result** | | | **The overall risk of bias** |
| 111,112 | Low | Low | Low | Low | Low | | | Low |



2

**Table 3. Meta-analyses and sensitivity analyses of single family rooms versus open bay units on infant outcomes**

| Outcome | Study populations (n), [references] | Total participants (n) | Participants in SFR (n) | Participants in OBU (n) | Heterogeneity $I^2$ (%) | | | |
|---|---|---|---|---|---|---|---|---|
| **Primary outcome** | | | | | | | | |
| | | | | | | *MD* | *95%CI* | *P-value* |
| Long term neurodevelopment* [cognitive] | 3 [115,124,135] | 680 | 330 | 350 | 42% | +1·42 | -1·11, 3·95 | 0·27 |
| Long term neurodevelopment* [cognitive] *Omitting*[135] | 2 [115,124] | 464 | 207 | 257 | 71% | +1·04 | -3·45, 5·52 | 0·65 |
| **Secondary outcomes** | | | | | | | | |
| *Continuous outcomes* | | | | | | *MD* | *95%CI* | *P-value* |
| Length of stay (days) | 12 [111,113–115,118,120,121, 124,125,128,130–132] | 4,702 | 2,298 | 2,404 | 75% | -1·33 | -2·77, 0·11 | 0·07 |
| Length of stay (days) *Only studies with low/moderate RoB* | 6 [111,115,120,124,128,130] | 3,863 | 1,900 | 1,963 | 82% | -2·46 | -5·89, 0·97 | 0·16 |
| *Dichotomous outcomes* | | | | | | *RR* | *95%CI* | *P-value* |
| Sepsis *As defined by authors* | 9 [111,115,117,120,121,124,125,128,130] | 4,165 | 2,055 | 2,110 | 0% | 0·63 | 0·50 – 0·78 | <0·0001 |
| Sepsis *Only culture proven sepsis †* | 6 [111,115,121,124,128,130] | 3,968 | 1,933 | 2,035 | 0% | 0·62 | 0·48 – 0·80 | 0·0002 |
| Sepsis *Per 1,000 patient days* | 9 [111,115,117,120,121,124,125,128,130] | NA | 1.95 | 2.92 | 4% | 0·67 | 0·51 – 0·87 | 0·002 |

**Table 3. Meta-analyses and sensitivity analyses of single family rooms versus open bay units on infant outcomes (continued)**

| Outcome | Study populations (n), [references] | Total participants (n) | Participants in SFR (n) | Participants in OBU (n) | Heterogeneity $I^2$ (%) | | | |
|---|---|---|---|---|---|---|---|---|
| Sepsis<br>*Only studies with low/moderate RoB* | 6 [111,115,120,124,128,130] | 3,859 | 1,895 | 1,964 | 0% | 0·65 | 0·50 – 0·85 | 0·001 |
| Breastfeeding at discharge *(any)* | 7 [112,114,116,117,124,130,131] | 1,109 | 499 | 610 | 0% | 1·06 | 1·00 – 1·13 | 0·04 |
| Breastfeeding at discharge *(any)*<br>*Only studies with low/moderate RoB* | 4 [112,116,124,130] | 907 | 407 | 500 | 18% | 1·03 | 0·91 – 1·16 | 0·63 |
| Breastfeeding at discharge *(exclusive)* | 5 [114,117,120,131,132] | 484 | 266 | 218 | 0% | 1·31 | 1·07 – 1·61 | 0·01 |
| Breastfeeding at discharge *(exclusive)*<br>*Only studies with low/moderate RoB* | 1 [120] | 99 | 68 | 31 | NA | 2·74 | 1·04 – 7·21 | 0·04 |
| Mortality | 7 [111,114,115,121,124,128,130] | 4,816 | 2,414 | 2,402 | 0% | 1·00 | 0·79 – 1·28 | 0·99 |
| BPD | 5 [111,115,121,124,128] | 3,800 | 1,849 | 1,951 | 42% | 1·03 | 0·76 – 1·38 | 0·87 |
| ROP | 5 [111,115,121,124,128] | 4,350 | 2,207 | 2,143 | 0% | 0·66 | 0·42 – 1·04 | 0·08 |
| IVH | 5 [111,115,121,124,128] | 4,348 | 2,207 | 2,141 | 0% | 1·03 | 0·70 – 1·51 | 0·88 |

* as measured by the BSID-III: Bayley Scales of Infant and Toddler Development, †: see Appendix, for definition of sepsis, 95%CI: 95% confidence interval, BPD: bronchopulmonary dysplasia, IVH: intraventricular hemorrhage, MD: mean difference, NA: not applicable, OBU: open bay unit, OR: odds ratio, RoB: Risk of Bias, ROP: retinopathy of prematurity, SFR: single family room

Chapter 3

# **Family integrated care in single family rooms for preterm infants and late-onset sepsis:** a retrospective study and mediation analysis

*Pediatric Research 2020; 88: 593-600*

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# ABSTRACT

### Background

During hospital stay after birth, preterm infants are susceptible to late-onset sepsis (LOS).

### Objective

To study the effect of family integrated care in single family rooms (SFR) compared to standard care in open bay units (OBU) on LOS. Peripheral- or central-venous catheters (PVC/CVC) and parenteral nutrition (PN) were investigated as potential mediators. Secondary outcomes were length of stay, exclusive breastfeeding at discharge and weight gain during hospital stay.

### Methods

Single-center retrospective before-after-study with preterm infants admitted ≥3 days.

### Results

We studied 1,046 infants (468 in SFR, 578 in OBU, median gestational age 35 weeks). SFRs were associated with less LOS (adjusted OR 0.486,95%CI 0.293;0.807,p=0.005). PVC (indirect effect -1.757, 95%CI -2.738;-1.068), CVC (indirect effect -1.002,95%CI -2.481;0.092) and PN (indirect effect -1.784,95%CI -2.688;-1.114) were possible mediators of the effect. PN was the main mediator of the effect of SFR on LOS. We found shorter length of stay (median in SFR 10 days (IQR:5-24) and in OBU 12 days (IQR:5-22), adjusted beta -0.088 95%CI: -0.159;-0.016, p=0.016), but no differences in weight gain or exclusive breastfeeding at discharge.

### Conclusion

SFRs were associated with decreased incidences of LOS and shorter length of hospital stay. The positive effect of SFRs on LOS was mainly mediated through a decreased use of PN in SFRs.

# INTRODUCTION

## Background

Each year approximately 15 million children are born preterm, and the largest proportion (85%) is born moderate or late preterm (between 32 and 37 weeks of gestation).[8] Moderate or late preterm infants can also spend a considerable time in the neonatal intensive care unit (NICU) after birth and they are susceptible to late-onset sepsis (LOS, occurring $^{3}$72 hours after birth).[145] LOS has a multifactorial origin, and risk factors for LOS include biological immaturity of immunological defense mechanisms, frequent disruptions of natural barriers (skin and mucous membranes), the presence of peripheral or central venous catheters, poor hygienic procedures or prolonged and widespread initial empirical antibiotic treatment.[97,146]



In the technological environment of the modern day NICU parents cannot always be present and emotional and physical closeness is impaired as preterm infants are cared for in open bay units .[49] The European Standards of Care for Newborn Health recommend that each unit should strive for an optimal design to support family-centered care, thereby facilitating parent participation in care and optimizing outcomes (including LOS) in this vulnerable patient population.[76,77] Supporting these recommendations, a recent meta-analysis (including 9 studies and 4,165 patients) revealed that hospitalizing preterm infants in single family rooms (SFR) was associated with a reduced incidence of LOS during hospital stay relative to OBU.[147]

However, in these previous studies, the mechanisms underlying the decrease in LOS events has remained unidentified and the participation and presence of parents and the association with infections remains unclear.[148,149] Specifically, no studies have described the association between the use of peripheral intravenous catheters (PVCs), central venous catheters (CVCs) or parenteral nutrition (PN) and the incidence of LOS when infants are cared for in different environments.

In 2014, our neonatal level 2 department was rebuilt to SFR, thereby allowing parents to be present 24 hours per day, with complete couplet care for mother-infant-dyads.[150] Also, we simultaneously started a complementary family integrated care (FICare) program.[55] In this program, parents provide most of the care for their infants, they are invited to participate in family-centered rounds and have educational group sessions to learn about various aspects of prematurity.

### Objective

We studied the effect of hospitalizing preterm infants in single family rooms with a complementary family integrated care program compared to standard care in open bay units on the incidence of late-onset sepsis in our level 2 neonatal ward. Additionally, we studied peripheral venous catheters, central venous catheters, and parenteral nutrition as potential mediators in the pathway between single family rooms and late-onset sepsis. Secondary outcome measures were length of hospital stay, exclusive breastfeeding at discharge and weight gain during hospital stay.

## METHODS

We used the Transparent Reporting of Evaluations with Nonrandomized Designs (TREND) checklist[151] and the Strengthening the Reporting of Observational Studies in Epidemiology for Newborn Infection (STROBE-NI) case-ascertainment methods.[152]

### Study design

This was a single center retrospective study, with a before-after design. We studied preterm infants admitted during the period of standard neonatal care in OBU (January 2012 through June 2014) versus preterm infants admitted to FICare in SFR (January 2015 through December 2016). An information specialist, independent of the objective of this research was asked to provide all cases of preterm infants admitted to the hospital during the study period. Data collectors were blinded to the objective of this research and collected data of all preterm infants from January 2012 through December 2016. Due to the construction of the new ward, FICare implementation and training of health care professionals, data between June 2014 and January 2015 were not used for this study. Data was collected from medical records during the years 2017 and 2018.

### Population

Eligible patients were all infants born in or transferred to the level 2 neonatal ward in a teaching hospital in Amsterdam, the Netherlands. Preterm infants (<37 weeks' gestation) with a length of hospital stay $^3$3 days were included. For a detailed explanation on the population of preterm infants studied and levels of neonatal care in the Netherlands, see the Appendix.

### Intervention

#### Family Integrated Care (FICare)

In the FICare model parents were trained to be the primary caregiver of their infant, and nurses supported, instructed and counseled parents. Parents were invited but not obligated to be present $^{3}$8 hours per day, and rooming-in facilities were present if they wanted to stay during the night. Parents were actively encouraged to participate in their infant's care by providing feedings by nasogastric tube, breast or bottle, providing skin-to-skin care (by mothers and fathers), weighing and regulating temperature control.

Family-centered rounds were implemented and included active parental participation in medical decision making on daily medical rounds and involving them in the process of patient management together with the nurses and doctors.[153,154] Weekly, parents had group-sessions to learn and talk about prematurity and their infant's hospital stay, guided by health care professionals or veteran parents.[55]



Nurses provided cardiorespiratory monitoring as well as treatments such as intravenous fluids or antibiotics, placing nasogastric tubes, providing respiratory support and phototherapy.[55,89,155]

### Single Family Rooms

The new neonatal, maternity and obstetric ward of the hospital allowed mothers and their infants to always stay together in one family room. They were never separated, even when one or both needed medical care (providing complete couplet-care, Appendix Figure S1).[150] Other family members could also be present 24 hours per day. With this new architectural design of the mother-child-center, maternity and neonatology services were fully integrated, with trained professionals with special skills to provide simultaneous medical care for ill mothers and/or preterm infants.

## Control/reference treatment

### Standard Neonatal Care in Open Bay Units

Standard neonatal care in OBU was provided before October 2014. In this ward infants stayed together in an open bay ward (a maximum of 18 infants in the ward), with incubators and beds lined up next to each other separated by curtains (Appendix Figure S2). The OBU was close to, but physically separated from the maternity ward. Parents could visit their child, participate in routine infant care, and provide skin-to-skin contact. Due to the setting, the duration mothers and fathers could stay at the bedside of their infant was limited. Rooming in facilities were not present.

During the hospital stay of their infant, parents were stimulated by the nurses to take part in the basic care of their infant. Medical rounds were done in a separate room from the OBU, attended by the nurses and the doctors, without the parents. Approximately two days before discharge, parents could room-in with their infant in a family room near the neonatal ward.

### Inclusion- and exclusion criteria

Infants were included if they were born between 24 and 36 6/7 weeks' gestation, with a post-conceptional age <44 weeks on admission and if they had a length of hospital stay in the level 2 ward of $^{3}$3 days. Infants were excluded if they were born abroad (not in the Netherlands) or had a congenital anomaly (for example severe congenital heart-defects requiring surgery or Down's syndrome).

### Case-ascertainment

Late-onset sepsis was defined as a clinical suspicion of sepsis with the physician's decision to do diagnostic testing because of symptoms of illness, to examine and culture bodily fluids (blood, urine and cerebrospinal fluid) for the presence of bacterial micro-organisms and to start antibiotics. The clinical signs should present between $^{3}$72 hours and 90 days after birth, during hospitalization for prematurity. The local hospital protocol defined the following signs to be present to consider a bacterial infection in the infant: increased, decreased, or instable temperature, respiratory problems including tachypnea, nasal flaring, grunting, labored breathing, cyanosis and apneas. Other possible signs were circulatory instability, feeding problems (feeding intolerance and vomiting), lethargy or irritability.

The following definitions were used:

*Total group of LOS:* If signs were present of LOS, cultures were drawn, and antibiotics were started.

*Group 1: Culture proven LOS*: When cultures were positive, independent of the duration of treatment for sepsis.

*Group 2: Clinical suspected but not confirmed LOS:* If diagnostics were done, and the antibiotic treatment was given for a maximum of 3 days with negative cultures.

*Group 3: LOS treated $^{3}$7 days with antibiotics:* When the infant was treated with antibiotics $^{3}$7 days after clinical signs of LOS, and cultures were negative.
We defined LOS as probable and treated with infants either in *Group 1* or *Group 3*.

A minimum of 0,5 to 1 mL of blood was required for culture on clinical indication as defined by the local hospital protocol. Cultures were drawn before antimicrobial administration. We only considered the first LOS episode during hospital stay in the department for the primary outcome and did not analyze relapse episodes.

One author (AvK) reviewed cases of children who developed LOS within 3 days after transfer from the level 3 NICU. These cases were checked anonymously and blinded to their allocation to OBU or SFR for case ascertainment (assigned to LOS developed in our hospital, or LOS developed in the referring NICU). We calculated LOS events per 1,000 hospital days.

### Potential mediators

We considered PVCs, CVCs, and PN as possible mediators in the pathway between SFR and LOS. In infants that developed LOS, mediators were only coded as present (yes/no) if they were started up to 1 day before the development of LOS. If infants received PVCs, PN or CVCs after development of clinical signs of LOS, mediators were not coded as present. In children that did not develop LOS, mediators were coded as present ever (yes/no) during hospital stay. All potential were analyzed in simple mediation models.



Additionally, mediators were analyzed in multiple mediation models if they were identified as a mediator in the simple mediation model. As PN can be given over PVC or CVC, we researched two different models. In model 1 we investigated PVC (without PN), CVC (without PN) and PN. In model 2 we analyzed PVC (without PN), CVC (without PN) and PN over PVC, PN over CVC, or PN over both PVC and CVC.

### Secondary outcome measures

The secondary outcomes evaluated were rate of weight gain during hospital stay (average weight gain as calculated by Cormack[156], using the following formula as an indicator of child health during hospital stay: 1000x(ln(discharge weight/birthweight)/(date of discharge–date of birth), length of hospital stay (in days, from admission to discharge), and exclusive breastfeeding (100% breastfeeds or expressed breastmilk) during the 24 hours before discharge.[157]

### Sample size calculation

In a pilot study, we found LOS in approximately 10% of preterm infants admitted to our OBU. We hypothesized that FICare in SFR would reduce the incidence of LOS to 5%. With a power of (1- β) 80%, and a significance level of (α) 0.05, we needed 436 patients in OBU, and 436 patients in SFRs.

### Statistical analysis

For continuous variables we evaluated if these were normally distributed using histograms. If normally distributed, mean and standard deviation (SD) were calculated. If variables were non-normally distributed median and interquartile range (IQR) were calculated. For non-normally distributed data, we first applied a (natural) logarithmic- or square root transformation to obtain normal distribution. We performed an independent t-test or Mann-Whitney test as appropriate.

For binary variables (e.g. sex and singleton) number and percentages were calculated. We calculated the Chi-square ($\chi^2$), to test for differences. If expected cell-counts were less than 5, we calculated differences with the Fisher's exact test.

**Missing data**

For missing data the proposed guidance as explained by Sterne et al. was applied.[158] Little's MCAR test was used to test the missing completely at random assumption for continuous variables. We applied the multivariate imputation by chained equations (mice) procedure to missing data, with 30 imputations and 50 iterations to obtain a dataset for further analyses.[159] Pooled estimates were derived applying Rubin's Rules.[160] Convergence was checked graphically with iteration plots.

**Evaluation of the primary and secondary outcomes**

To evaluate the association between SFR and binary outcomes (late-onset sepsis, exclusive breastfeeding at discharge), we calculated odds ratios (ORs) with multivariable logistic regression analyses. For continuous outcome variables (length of stay and weight gain) multivariable linear regression analyses were performed. The crude regression models were adjusted for confounders identified in the literature or if the beta-regression coefficient differed at least 10% from the crude beta-coefficient. The following variables were assessed and included in the adjusted model: birthweight (in grams), post-intensive care status, sex and previous treatment with antibiotics for early onset sepsis (<72 hours after birth), and culture proven late-onset sepsis for secondary outcomes. If collinearity was present, the strongest confounder (with the largest change in crude beta-coefficient) was used to adjust for. For the difference in incidence of sepsis (per 1,000 hospital days) between SFR and OBU we calculated incidence rate ratios (IRRs).

**Mediation analysis**

Mediation analyses on the imputed dataset were applied to analyze, identify and explain the underlying mechanisms of the observed total effect of SFR on LOS (i.e. the *c*-path).[161] In addition to the total effect model two logistic regression models were fitted. In single mediator models, PVC, CVC and PN were included as individual potential mediators. In the first regression model the effect of SFR on the mediator was estimated (*a*-path). In the second regression model the effect of the mediator on LOS (*b* path) and the direct effect of SFR on LOS (*c'*-path) were calculated. Crude and adjusted mediation analyses were performed. In the adjusted analyses, confounders were added to all paths. We calculated the indirect effect as the product of the *a* and *b* coefficients. We used bootstrap 95% percentile confidence intervals based on 1,000 bootstrap resamples around the indirect effects.[162]

Additionally, we included all identified mediators in a multiple mediator model. Again, indirect effects were calculated as the product of the *a* and *b* coefficients. To summarize information provided by the multiple mediator models, we calculated proportions mediated. The proportion mediation was calculated by dividing the indirect effect of the separate mediators through the sum of the indirect and the direct effect.

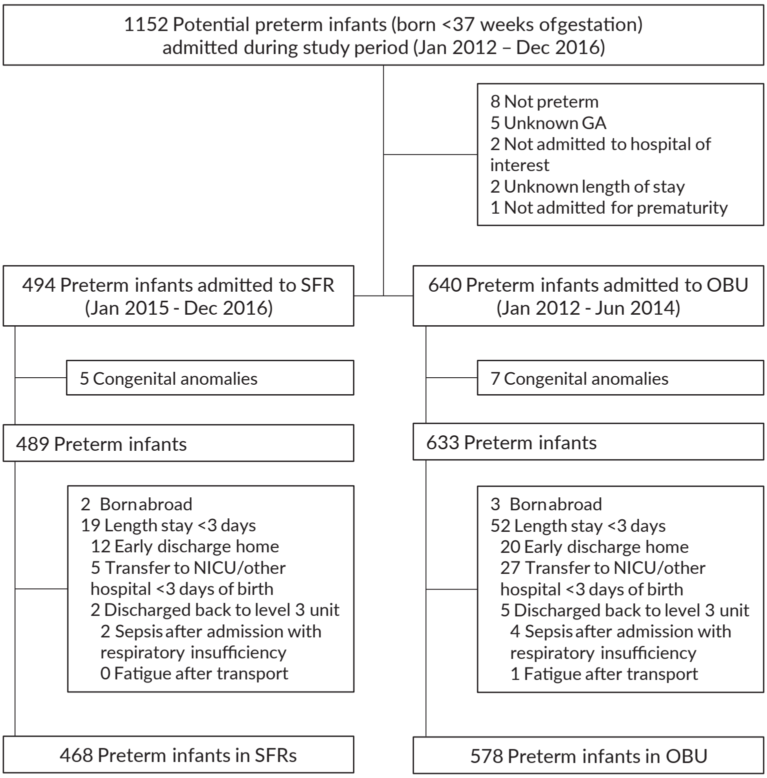
### Statistical packages and software

We used R for statistical analyses (version 3.6.1)[163] and specifically for multiple imputation the 'mice'-package[159], for analyzing missing data patterns the 'VIM'-package[164], the 'epiR'-package for the IRR[165], for Little MCAR's test the 'BaylorEdPsych'-package[166], and for the 95% percentile bootstrap confidence interval the 'boot'-package.[167] For all tests, a *p*-value of less than 0.05 was considered statistically significant.



## RESULTS

**Figure 1. Flowdiagram**




GA: gestational age, NICU: neonatal intensive care unit, SFRs: single family rooms, OBU: open bay unit

A total of 1,152 preterm infants were admitted during the entire study period. From this cohort, 1,046 preterm infants without exclusion criteria (468 in SFR and 578 in OBU) were admitted for $^{3}$3 days in our hospital (Figure 1).

The baseline characteristics of infants admitted to SFR or OBU are presented in Table 1. Infants admitted to SFR had a higher median gestational age than infants in OBU ($35^{+2}$ vs. $34^{+6}$ weeks respectively).

The proportion of males was larger in SFR (53.8% vs. 47.9%). Infants were more likely to be treated with antibiotics for early onset sepsis in the OBU group (21.2% vs. 14.5%). No differences were found in other baseline characteristics.

Fewer infants received PVC (31.2% in SFR vs 55.7% in OBU), CVC (2.4% vs. 6.9%) and PN (21.4% vs. 38.4%) during their stay in SFR compared to OBU (Table 2).

The Little MCAR test, revealed that data was not missing completely at random ($\chi^2$=130.9, p<0.00001) and 10 missing data patterns were present for continuous outcomes (and 36 if including all variables).

**Single family rooms and the association with late-onset sepsis**

We analyzed a total of 17,443 days of hospitalization (7,821 in SFR and 9,622 in OBU). Twelve children had a clinical suspicion of LOS within 3 days after transfer from another level 3 NICU. Eight events (4 in SFR and 4 in OBU) were assigned to the NICU of origin, and not included in the analyses. In SFR, 25 (5.3%) infants had a clinical suspicion of LOS and were started on treatment with antibiotics compared to 54 (9.3%) infants in OBU (adjusted OR 0.486, 95%CI 0.293; 0.807, p=0.005). Also, when analyzing this per hospital day it was lower in SFR than in OBU (3.2/1,000 vs. 5.6/1,000, IRR 0.570, 95%CI 0.340; 0.930, p=0.019). This association was also present for LOS treated for at least 7 days (Group 3). Though not significant, in SFR, infants were less often started on antibiotics for at least 2-3 days with negative cultures (adjusted OR 0.235, 95%CI 0.051; 1.082, p=0.063) and overall, less LOS events were probable in SFRs (adjusted OR 0.558, 95%CI 0.324; 0.961, p=0.036, Table 3).

On average, infants developed LOS symptoms and were started on antibiotics 6 days after hospital admission, and this was not different in the SFR and OBU group. In total, 42 infants had a culture proven LOS (Group 1), 17 (3.6%) in SFR and 25 (4.3%)) in OBU during hospital stay (adjusted OR 0.739, 95%CI 0.387; 1.410, p=0.348). The incidence in SFR was 2.2/1,000 compared to 2.6/1,000 hospital days in OBU (IRR 0.836, 95%CI 0.424; 1.611 p=0.570). If infants had a culture proven sepsis, sepsis was more often associated with a focal infection in SFR (14/17 (82%)) compared to OBUs (7/25 (28%)) (Appendix Table S2).

**Mediation analysis**

The relationship between SFR and LOS was mediated by PVC, CVC and PN individually, independent of the definition of LOS (Figure 2 and Appendix Table S3). Fewer infants received PVC (31.2% in SFR vs. 55.7% in OBU), CVC (2.4% vs 6.9%) and PN (21.4% vs 38.4%, Table 2) during their stay in SFR compared to OBU also when adjusting for confounding factors (*a* path, Figure 2 and Appendix Table S3). PVC, CVC and PN were positively associated with culture proven, LOS treated for 7 days and clinically suspected but not confirmed LOS (*b* path, Appendix Table S3).

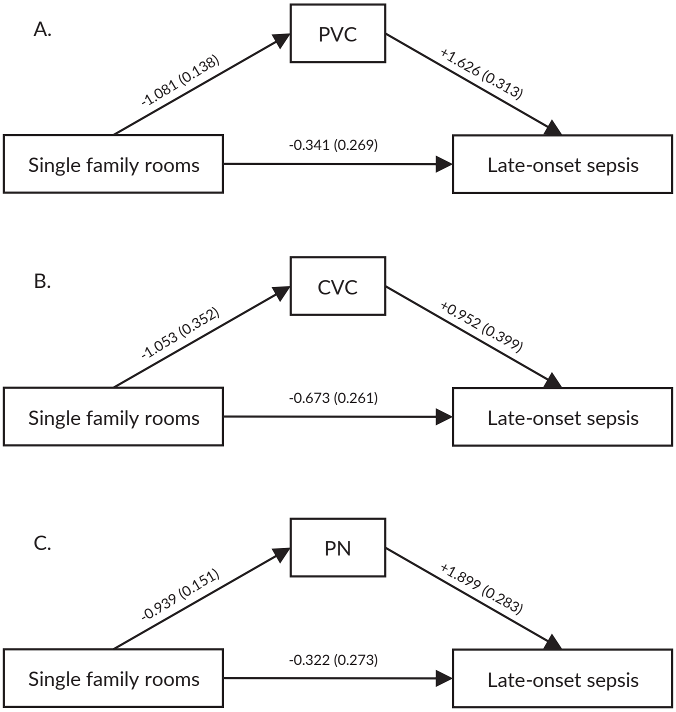


The indirect mediating effect of PVC and PN on LOS was present in single mediation analysis, also after adjusting for all possible confounders (indirect effect *(ab)*, Figure 2, Appendix Table S3).

The 95%-confidence interval of the indirect effect of CVC included zero in all analyses, indicating a non-significant indirect effect in the pathway between SFR and LOS. The total proportion of mediation through PVC, CVC and PN after adjustment for confounders was 83.7%, 59.8% and 84.7% respectively in the single mediation models (Appendix Table S3).

Within multiple mediation models (model 1) the effect of SFR on LOS was mainly mediated by PN and less through PVC without PN (Appendix Table S5, Appendix Figure S3). As none of the infants received CVC without PN, CVC in itself were not a mediator in the pathway between SFR and LOS (Appendix Table S4). In model 2, CVC without PN was not a mediator, PVC without PN was a moderate mediator and PN was a strong mediator of the effect of SFR on LOS. The 3 different pathways by which PN could be given, were all mediators of the effect of SFR on LOS (Figure 3). Within model 2, the effect of PN through PVC was stable over different definitions of LOS with narrow 95%-bootstrap confidence intervals of the indirect (*ab*) effect. PN over CVC, or PN over both were also mediators, but confidence intervals were very wide (Appendix Table S6).

**Figure 2. Single mediation models of FICare in single family rooms on late-onset sepsis.**




A: Peripheral venous catheters (PVC) as a mediator of the effect of FICare in single family rooms on late-onset sepsis in preterm infants.

B: Central venous catheters (CVC) as a mediator of the effect of FICare in single family rooms on late-onset sepsis in preterm infants.

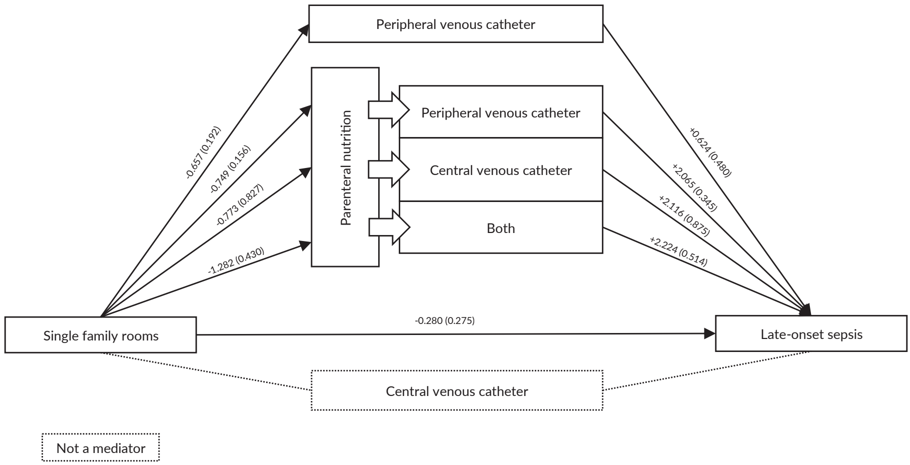
C: Parenteral nutrition (PN) as a mediator of the effect of FICare in single family rooms on late-onset sepsis in preterm infants.

All analyses are adjusted for birthweight, post-intensive care status, previously treated for early onset sepsis and sex.

## Single family rooms and the association with length of stay, exclusive breastfeeding at discharge and weight gain

The median length of stay in SFRs was 10 days (IQR 5 to 24) and in OBU 12 days (IQR 5 to 22). After adjusting for confounding factors, SFR was associated with a shorter length of stay (adjusted beta after natural logarithmic (ln) transformation; -0.088, 95%CI -0.158; -0.016, p=0.016). After adjusting for confounders, SFR was associated with higher exclusive breastfeeding at discharge and weight gain, but this was statistically not significant (Appendix Table S7).

**Figure 3. Multiple mediation model of FICare in SFR on late-onset sepsis**




Analysis is adjusted for post-intensive care status, previous treatment with antibiotics for early onset sepsis, birthweight and sex.

# DISCUSSION

## Key findings

In this study, peripheral and central venous catheters and parenteral nutrition were potential mediators in the pathway between single family rooms and late-onset sepsis. The positive effect of single family rooms on late-onset sepsis was mainly mediated through a decreased use of parenteral nutrition in single family rooms, and – to a lesser extent – decreased use of peripheral venous catheters.

These findings are consistent with our previous systematic review of SFRs and the incidence of LOS.[147] We show similar results in the rate of clinically suspected LOS events and proven LOS events per 1,000 hospital days. Also in line with previous research, PVC and CVC use in our population was associated with LOS[97], and we found a strong association between PN and the development of LOS.[97,168,169] Due to the retrospective nature of our study, we did not culture the PN for contamination and therefore it remains unclear if the PN itself caused the LOS, or if PN predisposed for LOS. Interestingly, even though lower usage of PN was present, no differences in weight gain was measured between the two groups.

The reason for the decreased use of PVC, CVC and PN in SFR remains to be further elucidated. Also, after correcting for gestational age, birthweight and previous treatment for early onset sepsis, this lower usage of catheters and PN was present.

Previous research has shown that parents are often stressed and concerned about painful procedures in their infant.[170] Parents were often more present during hospital stay in SFR, participated in family-centered rounds and aided in the caregiving and decision making in this vulnerable patient population.[154,171,172] They could have gathered more knowledge in the education sessions and had more parenting experiences during hospital stay with their infant.[55,155] One could consider that family-centered rounds invited to more elaborate discussions with the parents on the necessity of the use of catheters and PN.[171] Previous research, and observations from our research group showed that, if parents were present in SFR, clinicians were more likely to discontinue after multiple attempts to start interventions, including PN and catheters, despite the local hospital protocol.[134,173] As this was not measured in this retrospective study this should be addressed in future research.

In previous studies, parental participation was a mediator of the effect of SFR on clinical outcomes in children, including weight gain and short- and longterm neurodevelopment in previous studies.[134,135,172] One could consider that SFR facilitated parental presence and participation in our population as well. Parents could have provided health care professionals with important information on the clinical condition of their infant, and might have aided physicians in their suspicion and/or diagnosis of LOS (through their parental experience and knowledge of their child's normal behavior).[55,155,174] This could have resulted in less use of antibiotics in our population, and also this is reflected by the higher percentage of culture proven LOS in the SFR infants when compared to OBU infants. This might possibly represent the difference in reading clinical cues between parents and professionals, as symptoms of LOS can sometimes be difficult to interpret.[98] Obviously, the role of the parents and subsequent actions of the health care professionals in these SFRs, and the effect on the use of PN, catheters and (diagnosis of) outcomes needs to be explored in future (preferably prospective) research.

In line with previous studies we found that SFR is associated with a shorter length of stay.[111,175] This is specifically important to healthcare policy makers, as costs per patient are high.[176] Decreasing length of stay in large proportions of patients can save money and support the decision of hospitals to redesign their hospitals to single family rooms. However, this should be more elaborately studied in cost-effectiveness analyses.

## Strengths

For the first time, we have done mediation analyses on the effect of an intervention on the usage of intravenous catheters and PN and the outcome LOS in this vulnerable patient group. Mediation analysis provides insight in the mechanisms underlying the effect of SFR on LOS and therefore helps explain the differences observed in LOS between SFR and OBU.

In contrast to previous studies concerning this area of research, we have transparently shown where the data is missing, and how we dealt with this.[147] Multiple imputation has been shown to be robust when dealing with missing data.[159] As missing values are a problem in bootstrapping, and may lead to biased outcomes, multiple imputation also accounted for this.[162] The non-imputed outcome was more extreme (association of SFR with LOS; adjusted OR 0.378, 95%CI 0.198;0.688,p=0.002), and therefore the imputed dataset could give a more realistic measure of association.



As the information specialist and data-collectors were blinded to the objective of this study, selection bias into the study was minimized. Also, as we asked a researcher to classify infants developing LOS in our facility <3 days after transfer from the NICU, independent and blinded to group assignment, misclassification of LOS was minimized.

**Limitations**

As randomization was not possible due to the construction of the hospital, the non-randomization of this study should be considered when studying the associations between SFR and LOS. A possible explanation for a decreased incidence of LOS and decreased use of PN or PVC and CVC could also be the effect of time itself. As we studied two different time periods (before and after start of SFR), it might be possible that over time we reduced the use of PVC, CVC and PN or became more conservative in starting and/or more liberal in discontinuing antibiotics, which would be in concordance to previous studies [177]. Preferably randomized trials should be conducted to study this more thoroughly. One randomized trial did find that LOS was (non-significantly) decreased, however no information was given on the use of PVC, CVC or PN.[111]

Due to the high proportion of late preterm infants (≥35 weeks' gestation) in this study, we were unable to provide the age of full enteral or oral feedings in our population, as the volume of feedings often did not meet the criteria at time of discharge (volume of feedings >130ml/kg/day). For the subpopulation of children born <35 weeks of gestation in our facility (72 in SFR and 143 in OBU), the median number of days to full enteral feedings and removal of nasogastric tube was not significantly different (p=0.107, and p=0.327 respectively).

In absence of consensus on the quantification of breastfeeding and breastmilk rates, and due to the retrospective nature of our study, we used a previous reported measure for breastfeeding rates.[157] Future research should prospectively document and analyze breastfeeding rates with a standardized and validated scoring system that preferably also incorporates the quality and efficiency of breastfeeding preterm infants.[178]

Other etiological factors that might have reduced the need for catheters and PN during hospital stay, including feeding (in)tolerance and frequency, parental presence and participation, and duration of skin-to-skin-care during hospital stay were not measured in this study.[72] Also, we did not include other predisposing and possible mediators for LOS, for instance the amount of skin-breaks (for blood sampling or venous catheters), compliance to (hand)hygiene protocols and the cumulative amount of human milk given during hospital stay.[97,146] Future studies should include these variables in their analyses.

**Practice implications**

Our study shows that FICare in SFRs is associated with less late-onset sepsis events and a shortened hospital stay for preterm infants after birth, which can reduce costs. Also, we found less use of catheters and parenteral nutrition during hospital stay, which potentially explained the decrease of infections in our department. Our study therefore supports the development of SFR and implementation of FICare for the neonatal ward and can help policy makers and health care professionals decide on how to (re)construct hospital wards in the future and specifically level 2 neonatal wards. The population we studied, was a relatively healthy level 2 neonatal population, with concurrent low incidences of LOS in the two different environments. This study is applicable for level 2 neonatal wards, and specifically for those infants admitted for at least 3 days in a facility that is situated in a developed country such as the Netherlands.

This study is also meaningful for hospitals which may not be able to reconstruct their facilities to single family rooms, as they might be able to consider more discretely the need for PN, PVC, and CVC in the care of preterm infants.

# CONCLUSION

This study contributes to existing knowledge on the beneficial effect of family integrated care and single family rooms in preterm infants. We identified less late-onset sepsis events a shorter hospital stay and less use of peripheral and central venous catheters and parenteral nutrition in preterm infants admitted to single family rooms with complementary family integrated care program, with similar weight gain. By using mediation analysis, we identified that the reduction in late-onset sepsis in single family rooms was mediated by a reduced use of intravenous catheters and parenteral nutrition.

**Table 1. Baseline characteristics.**

| Characteristic | SFR infants (n= 468) | Missing (n (%)) | OBU infants (n=578) | Missing (n (%)) |
|---|---|---|---|---|
| Gestational age (wks$^{+days}$, median (IQR), range (min to max)) | $35^{+2}$ ($32^{+2}$-$36^{+2}$)*<br>$24^{+2}$ to $36^{+6}$ | 0 | $34^{+6}$ ($32^{+6}$-$36^{+1}$)*<br>$24^{+2}$ to $36^{+6}$ | 0 |
| Birthweight (grams, mean (SD)) | 2145 (708) | 0 | 2143 (656) | 1 (0.2) |
| Male (n (%)) | 252 (53.8) | 0 | 277 (47.9) | 0 |
| Apgar score at 1 min (median (IQR)) | 9 (7-9) | 4 (0.8) | 8.5 (7-9) | 8 (1.4) |
| Apgar score at 5 min (median (IQR)) | 9 (8-10) | 3 (0.6) | 9 (8-10) | 9 (1.6) |
| Inborn (n (%)) | 313 (66.9) | 0 | 415 (71.9) | 1 (0.2) |
| Post Intensive care status (transferred from level 3 NICU) (n (%)) | 146 (31.2) | 0 | 153 (26.4) | 1 (0.2) |
| Received surfactant previously | 40 (8.5) | 1 (0.2) | 54 (9.3) | 0 |
| Maternal age (yrs, mean (SD)) | 33.3 (4.7) | 4 (0.8) | 33.3 (5.2) | 8 (1.4) |
| Primipara (n (%)) | 251 (53.6) | 0 | 337 (58.0) | 1 (0.2) |
| Singleton (n (%)) | 322 (68.8) | 0 | 392 (67.8) | 0 |
| Vaginal delivery (n (%)) | 277 (59.2) | 0 | 333 (57.6) | 2 (0.3) |
| Antenatal corticosteroids (n (%)) | 153 (32.7) | 1 (0.2) | 196 (33.9) | 6 (1.0) |
| Treated for early onset sepsis (n (%)) | 68 (14.5) | 82 (17.5) | 123 (21.2) | 85 (14.7) |
| Had culture proven LOS previously in referral center (n (%)) | 28 (6.0)* | 7 (1.5) | 20 (3.5)* | 12 (2.1) |

*$p<0.05$, IQR: Interquartile range, LOS: late-onset sepsis, n: number, NICU: neonatal intensive care unit, OBU: open bay unit, SD: standard deviation, SFR: single family rooms, wks: weeks, yrs: years.

**Table 2. Presence of mediators in infants with and without late-onset sepsis in different hospital environments**

| Mediator | SFR infants | | | | OBU infants | | | |
|---|---|---|---|---|---|---|---|---|
| | Total (n=468) | With LOS (n = 25) | Without LOS (n=443) | Missing (n,(%)) | Total (n=578) | With LOS (n = 54) | Without LOS (n=524) | Missing (n,(%)) |
| PVC | 145 (31.0) | 18 (72.0) | 127 (28.7) | 6 (1.3) | 322 (55.7) | 47 (87.0) | 275 (52.3) | 13 (2.2) |
| CVC | 11 (2.4) | 1 (4.0) | 10 (2.3) | | 40 (6.9) | 9 (16.7) | 31 (5.9) | |
| PN | 96 (20.5) | 15 (60.0) | 81 (18.3) | | 222 (38.4) | 44 (81.5) | 178 (34.0) | |

CVC: central venous catheter, PVC: peripheral venous catheter, n: number, LOS: late-onset sepsis, OBU: open bay unit, PN: parenteral nutrition, SFR: single family room

**Table 3. The association between single family rooms and late-onset sepsis in the imputed dataset**

| Outcome | SFR infants (n=468) | OBU infants (n=578) | OR | 95%CI | OR[a] | 95%CI[a] |
|---|---|---|---|---|---|---|
| Overall LOS (*Total group*), n (%) | 25 (5.3) | 54 (9.3) | 0.548 | 0.335; 0.895* | 0.486 | 0.293; 0.807** |
| Culture proven LOS (*Group 1*), n (%) | 17 (3.6) | 25 (4.3) | 0.834 | 0.444; 1.564 | 0.739 | 0.387; 1.410 |
| Symptoms of LOS, maximum of 2-3 days of antibiotics, culture negative (*Group 2*), n (%) | 2 (0.4) | 11 (1.9) | 0.221 | 0.049; 1.005 | 0.235 | 0.051; 1.082 |
| LOS treated $^{3}$7d of antibiotics, culture negative (*Group 3*), n (%) | 6 (1.3) | 18 (3.1) | 0.404 | 0.159; 1.027 | 0.337 | 0.133; 0.907* |
| LOS probable and treated (*Group 1 and Group 3*), n (%) | 23 (4.9) | 43 (7.4) | 0.643 | 0.381; 1.084 | 0.558 | 0.324; 0.961* |

*p<0.05, **p<0.01, [a]Adjusted for post-intensive care status, previous treatment with antibiotics for early onset sepsis, birthweight, and sex, 95%CI: 95% - confidence interval, AB: antibiotics, d: days, LOS: late-onset sepsis, NA: not applicable, OBU: open bay unit, OR: odds ratio, SFR: single family room

Chapter 4

# **Hospitalising preterm infants in single family rooms versus open bay units:** a systematic review and meta-analysis of impact on parents

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# ABSTRACT

### Background

Many parents develop stress-related symptoms and depression when their preterm infant is hospitalised in the neonatal intensive care unit(NICU) after birth. We reviewed the evidence of parent well-being with preterm infants hospitalised in single family rooms(SFRs) or in open bay neonatal units(OBUs).

### Methods

For this systematic review and meta-analysis, we searched MEDLINE, EMBASE, PsycINFO, Cochrane Central Register of Controlled Trials(CENTRAL), Web of Science, Clinicaltrials.gov, and International Clinical Trials Registry Platform(ICTRP) databases from inception through 22 November 2019 using controlled terms and text words related to prematurity and NICU-design. We included randomised and non-randomised studies comparing outcomes in parents with preterm infants admitted to SFRs or OBUs. Methodological quality was assessed using Cochrane Collaboration's Risk of Bias Tool for randomised controlled trials and the Risk of Bias Tool for Non-Randomised Studies of Interventions(ROBINS-I). Outcomes included: parental stress, satisfaction, participation (presence/involvement/skin-to-skin care), self-efficacy, parent-infant-bonding, depression, anxiety, post-traumatic stress, empowerment, and degree of family-centred care. Summary estimates were calculated using random effects models with standardised mean differences(SMDs). PROSPERO registration:CRD42016050643.

### Findings

We identified 614 unique publications. Eleven study populations (1,850 preterm infants, 1,549 mothers and 379 fathers) were included. All but one study were at serious to critical risk of bias. SFRs were associated with higher levels of parental presence, involvement, and skin-to-skin care. Upon discharge, SFRs were associated with lower stress levels (n=828 parents, SMD-0·30,95%CI -0·50; -0·09, p<0·004, $I^2$=46%), specifically NICU-related stress (n=573, SMD-0·42,95%CI -0·61; -0·23, p<0·0001, $I^2$=0%). In majority of studies higher levels of empowerment, family-centred care, and satisfaction was present with SFRs. No differences were found for anxiety, parent-infant bonding, or self-efficacy. Depression was high (up to 29%) but not different between settings. No studies described post-traumatic stress.

### Interpretation

Single family rooms seem to facilitate parental presence, involvement, skin-to-skin care, and reduce NICU-related parental stress.

# INTRODUCTION

Every year, 14·9 million infants (approximately 11% of all livebirths worldwide) are born preterm, and this number is rising.[8] After birth, preterm infants can spend a considerable amount of time in the neonatal intensive care unit (NICU) before discharge home. This period can be very stressful for parents.[60] Parental stress arising from the experience of the NICU is an important issue that potentially impacts parenting behaviour and long-term emotional and health problems in parents and their infants.[62] Currently, most preterm infants are hospitalised in open bay units (OBUs), with clusters of infants on the same ward and limited accommodations for parents to be present continuously with their infant. The physical setting of the OBU potentially limits the emotional and physical closeness between parents* (caregivers) and their infant.[49]



The recently published European Standards of Care for Newborn Health and the World Health Organisation Survive and Thrive report recommend to accommodate parents in skin-to-skin care (SSC), to actively welcome and engage parents in the care of their newborn, and to facilitate parental presence throughout the 24 hours by an optimal design of the NICU.[75–78]

More and more NICUs are building single family rooms (SFRs) to accommodate parents to be present continuously during the day and at night with their infant. A previous systematic review and meta-analysis showed lower incidences of sepsis and increased exclusive breastfeeding rates upon discharge and no difference in long-term neurodevelopment for preterm infants hospitalised in SFRs compared with OBUs.[147] Another systematic review showed parents to experience increased privacy and feeling like a family-unit in SFRs compared to OBUs.[101] However, the impact of SFRs on well-being of parents of preterm infants has not been assessed before.

In this systematic review and meta-analysis, we reviewed the evidence on whether the physical design of a NICU has an impact on the well-being of parents of preterm infants and their participation during infant hospital stay. We compared outcomes of parents of preterm infants hospitalised in either SFRs or OBUs.

# METHODS

### Search strategy and selection criteria

This systematic review and meta-analysis used the same methods as our previous paper on infant outcomes.[147] A full protocol was published before conducting this research,[103] and parental outcomes were prespecified to be secondary outcomes.

We used the Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA-)guidelines.[102] An information specialist (JL) performed a broad search (adapted from the initial search)[147] in MEDLINE, EMBASE, PsycINFO (all via the OVID interface), the Cochrane Central Register of Controlled Trials (CENTRAL), Web of Science, Clinicaltrials.gov, and the International Clinical Trials Registry Platform (ICTRP) databases from their inception through 22 November 2019 (Appendix). There were no restrictions to language, date, study type, or publication status. We cross-checked reference lists and citing articles of identified relevant papers. We required studies to compare well-being of *parents* of preterm infants admitted to SFRs or to OBUs and to provide summary estimates of outcomes in parents.[103] We included randomised and non-randomised clinical trials. Two researchers (NRvV and SRDvdS) independently screened abstracts and assessed full-text articles for inclusion.

**Data analysis**

We calculated kappa and specific agreement for screening of studies. We collected the data as described in the protocol.[103] We contacted study authors up to twice to clarify (missing) data in included and potentially eligible studies. We defined a population as parents of infants from the same hospital during the same time-period of study. Two investigators (NRvV and SRDvdS) applied the Cochrane Collaboration's Risk of Bias Tool for randomised controlled trials and the Risk of Bias in Non-Randomised Studies of Interventions (ROBINS-I) tool to each study, at the study level, separately and independently.[105] Two investigators (NRvV and SRDvdS) independently examined the questionnaires and outcome measures used in the studies and grouped them into discrete conceptual categories (Appendix). Discrepancies were resolved via discussion within the research team. The prespecified outcomes included parental stress, satisfaction, participation, self-efficacy, and parent-infant bonding. Parent participation was further defined as: *presence* (amount of time parents are physically present with the infant in the hospital during hospital stay of the infant), *involvement* (amount (of time) parents are taking part in the care of their infant), and *skin-to-skin care* (amount of time parents provide SSC to their infant). Outcomes of additional relevance included during the review process were depression, anxiety, post-traumatic stress, empowerment, and degree of family-centred care (FCC).

We used Review Manager (version 5.3; the Cochrane Collaboration) and the 'meta' and 'metafor' packages in R (version 3.6.1) to conduct meta-analyses, sensitivity and subgroup analyses.[110,179] We assessed heterogeneity using the $I^2$ test for heterogeneity. We used a random-effects model for meta-analysis if heterogeneity was assessed to be acceptable ($I^2 \leq 50\%$) on crude estimates. In case of substantial or considerable heterogeneity ($I^2 > 50\%$) no pooled results were reported. Continuous data were analysed by computing the standardised mean difference (SMD) with 95% confidence intervals (CI) if studies assessed the same construct with different measurement instruments.[180] We calculated means and variances if they were not reported in the original publication as proposed previously.[108] We performed sensitivity analyses to estimate the effect of different assumptions on outcome variables (prespecified were risk of bias (RoB), gestational age (GA), and start of SFR care). Predefined subgroup analyses were performed for parent participation (analysing studies with higher levels of parent-involvement in SFR, significant more SSC in SFR, or >8 hours per day difference in parental presence between SFR and OBU). We added subgroup analyses on different constructs of stress (biomarkers, NICU-related parental stress, and parenting stress; Appendix). We prespecified to assess publication bias with funnel plots and to perform meta-regression analyses for outcomes assessed in more than 10 studies. This study was registered in PROSPERO (International Prospective Register of Systematic Reviews) on 2 November 2016 (registration number:CRD42016050643). Deviations from the protocol are described in the Appendix. This work was exempted from medical ethical approval as we used data from patients enrolled in studies and trials already approved by relevant ethical committees.



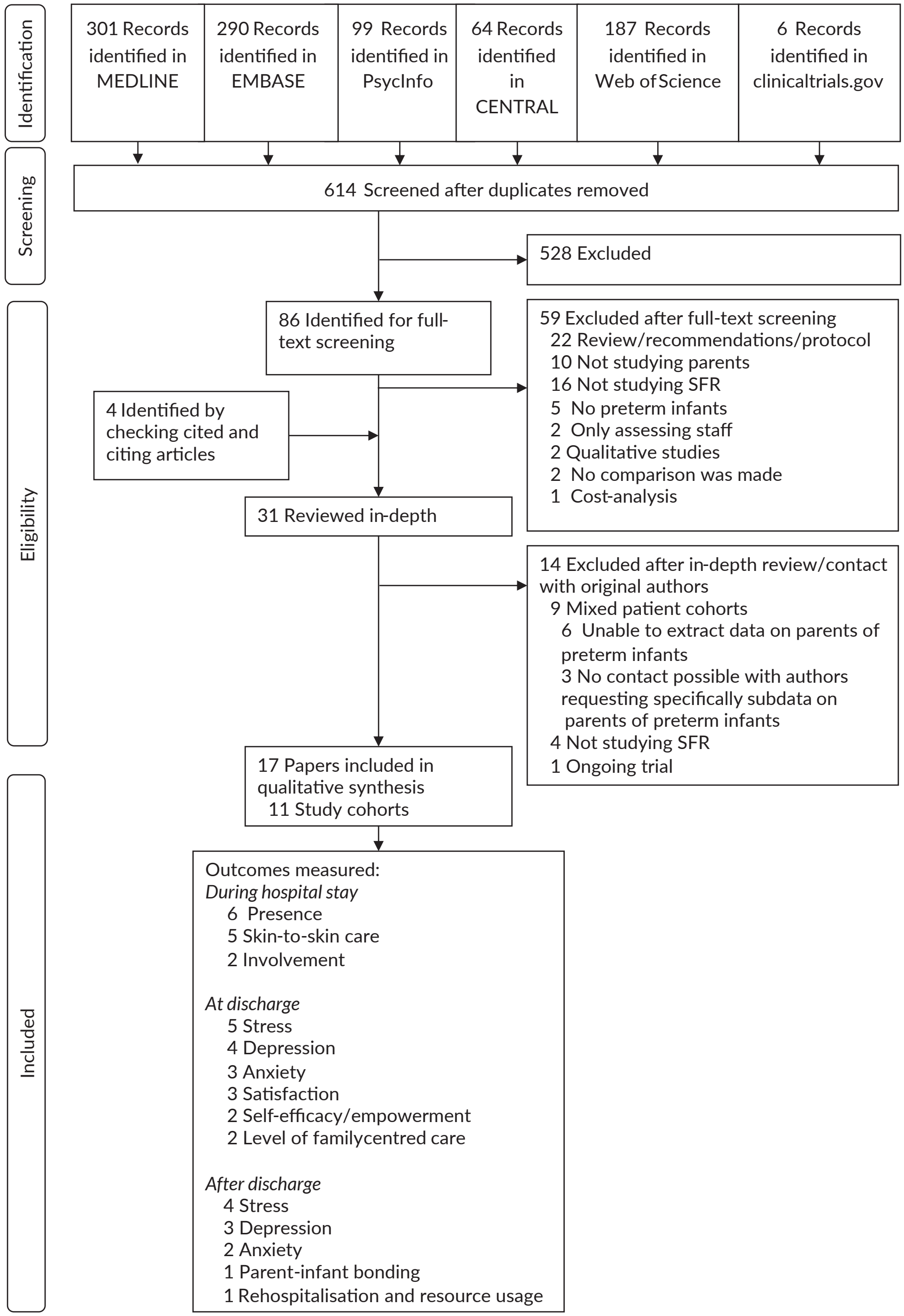
## RESULTS

A total of 614 records were identified in our search. Eighty-six references were identified for full-text screening (Figure 1). Thirty-one papers were reviewed in-depth. 24/27 (89%) of authors responded for additional information, and one full original dataset was provided.[131]

Eleven study populations (1,850 preterm infants, 1,549 mothers and 379 fathers) were included in 17 papers (Table 1). Seven papers provided information about fathers.[131,181–186] Five study populations were described in multiple papers (Table 1, for a detailed description see Appendix).[113,114,134,135,181–183,186,187] Care for mother and infant direct postnatally (couplet-care) was provided in 3 hospitals[118,186,187], and if mother was stable (usually 48 hours after delivery) in 1 hospital.[181–183] Facilities for parents to be present in the NICU were as described in the Appendix. The papers were published between 2004 and 2019, and all were performed in middle- to high-income countries.

Data pertaining to RoB assessments are listed in Table 2. The randomised clinical trial (RCT) was considered to be at some concerns on RoB, the RoB of all of the other studies ranged from serious to critical. One study provided insufficient information to assess RoB across at least one domain (see Appendix for a detailed explanation).[121]

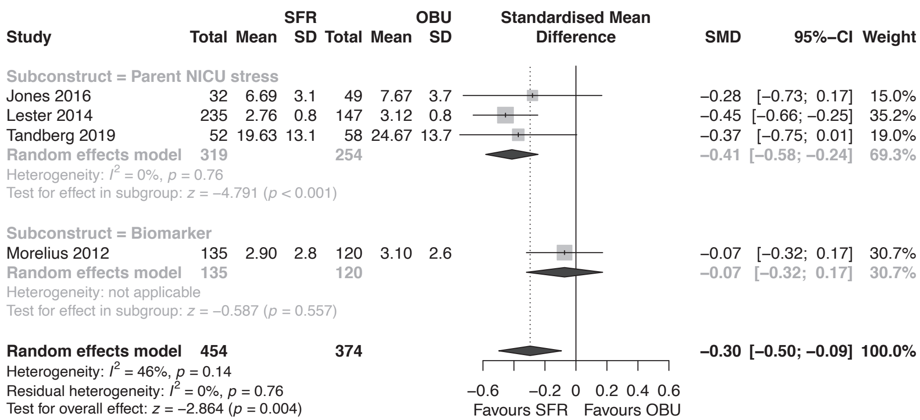
**Figure 1. Flowdiagram**



Including all studies that measured parental stress at discharge, statistical heterogeneity was high ($I^2$=79%, n=909 parents, Table 3) and no meta-analysis was performed.[112,116,131,134,135,183] Heterogeneity decreased when accounting for RoB (including only studies with low risk of confounding , $I^2$=63%, Appendix).[112,131,134,135], but not for GA[116,134,183] or start of SFR care at admission[116,131,134,183] (88% and 83%, respectively). One study specifically influenced heterogeneity, as it reported higher levels of stress on the parental role alteration *subscale* but not on all aspects of NICU-related parental stress.[116] Omitting this study decreased heterogeneity ($I^2$=46%) and SFRs were associated with lower parental stress (n=828,SMD-0·30,95%CI-0·50;-0·09,p<0·004). Also subgroup analyses on constructs of stress decreased heterogeneity: non-significant lower levels of salivary cortisol[112] and specifically less NICU-related parental stress was present in SFRs (n=595,SMD-0·41,95%CI-0·58;-0·25,p<0·001,$I^2$=0%,Figure 2). Two to four months after infant discharge, no differences in stress was found (see Table 3), all studies were at serious RoB and all infants were >32 weeks of GA.[113,118,131] No difference was noted analysing only infants admitted straight after birth to SFRs (Appendix).[118,131,183]



**Figure 2.**




SFR: single family room, SMD: standardized mean difference, OBU: open bay unit,

Statistical heterogeneity was high for anxiety at discharge ($I^2$=81%) and RoB serious in all but one study[131]. In this study significantly lower anxiety scores were found upon discharge (n=81,SMD-0·55, 95%CI-1·00;-0·10,p=0·02)[131]. Heterogeneity did not decrease when accounting for other aspects of RoB, GA or start of SFR care.[116,131] In one study (assessing state and trait anxiety), mothers in SFRs had higher trait anxiety, but experienced lower state anxiety than mothers in OBUs.[116] After discharge, parent anxiety did not differ between admission to either of two environments (n=136,SMD-0·17, 95%CI-0·51;0·17,p=0·316,$I^2$=0%).[131,183]

Up to 29% of parents had depressive symptomatology upon discharge. Heterogeneity between studies was high ($I^2$=87%, Table 3).[116,131,135,183] In sensitivity analyses of RoB, heterogeneity decreased ($I^2$=0%), and no statistically significant difference in parent depression upon discharge was noted (n=297,SMD-0·10,95%CI-0·33;0·14,p=0·42, Appendix).[131,135] Furthermore, no differences were found after infant discharge for parent depression (n=185,SMD-0·15,95%CI-0·48;0·18,p=0·372,$I^2$=16%,Table 3).[113,131] One study reported significantly lower depressive symptoms from admission up to 4 months for mothers with infants admitted to SFRs.[183]

During hospital stay, higher levels of empowerment in SFRs and more confidence in taking care of an infant without an attending staff member one week prior to discharge were reported.[117] Parents in SFRs reported feeling heard and receiving greater guidance and (emotional) support from nurses[182], and they rated the degree of FCC and satisfaction higher compared with parents in OBUs.[121,134,186] No differences in self-efficacy upon discharge were found.[131] After discharge, mothers reported less need for information about understanding their infant's behaviour and about breastfeeding[117]. Mothers in SFRs had fewer acute care visits with their infant and fewer telephone consultations with a physician after discharge.[114] No differences in parent-infant bonding during hospital stay and after discharge was found as they scored high in both environments.[183]

Parent participation was described in 15 studies (9 populations,Table 1,Appendix)[112–114,116,118,131,134,135,181,182,184–187] and measured in 8 populations.[113,114,116,118,131,134,135,181,182,184–187] It included reports on time of parental presence with the infant in the hospital[116,118,134,135,183,187], levels of parent-involvement in care[134,135,182,186,187] and amount of SSC.[116,118,134,135,183,187] No meta-analysis was performed, as statistical heterogeneity was high ($I^2$=98%, 78%, 96% respectively,Appendix).

Overall, in five out of six populations (n=486 parents in SFRs, n=404 parents in OBUs) increased parental presence was reported in SFRs.[114,116,131,134,135,181,187] In one study, lower presence in SFRs compared with OBUs was found if mother was not hospitalised.[113,114] Overall, parental presence in SFRs(range:3·6 to 22·4 hours per day) was higher than in OBUs(range:2·4 to 8·0 hours per day). When accounting for RoB (specifically confounding), heterogeneity decreased and parents were significantly more present in SFRs (n=417,SMD+0·59, 95%CI 0·36 to 0.83,p<0·0001,$I^2$=7%,Appendix)[131,134] Two studies in Sweden did not measure parental presence, but reported infants cared for with continuous SSC (24 hours per day) for the first week of life in SFRs[184] or parents expected to be with the infant around the clock.[112,184] If studies found >8 hours difference per day in parental presence between SFR and OBU, SFRs were associated with lower levels of parental stress and depression upon discharge (Appendix).[183]

The number of days per week that parents were involved was higher (4·5 days in SFRs versus 3·6 days in OBUs, without risk of confounding)[134,135], and parents participated more in discussions during the doctor's round in SFRs.[182]

Seeing or holding the infant skin-to-skin commenced earlier when infants were hospitalised in SFRs.[116,185] The amount of SSC was higher in SFRs (range: 1·9 to 24 hours per day) than in OBUs (range:0·5 to 2·5 hours per day,Appendix). Statistic heterogeneity was high ($I^2$=96%)[118,134,135,181,182,184,187], and did not decrease with sensitivity analyses. In studies with significant higher levels of SSC[134,135,183] SFRs were associated with significantly lower stress levels (n=492,SMD-0·44,95%CI-0·62;-0·25,p<0·0001,$I^2$=0 %,Appendix).[135,183]

In four study populations 379 fathers were present,[131,181–187] and 72 fathers were assessed on well-being (Appendix).[131,183] In one study[131] one father in an OBU had extremely severe depression upon discharge; no fathers in SFRs had depression symptomatology. Three fathers in OBUs had anxiety and stress upon discharge compared with one father in SFRs. In the other study (n=66), fathers had significantly lower stress levels in SFRs compared to OBUs, but no differences were noted on depression, anxiety, or parent-infant bonding.[183]

As none of the outcomes were assessed in more than 10 studies, meta-regression analysis and publication bias was not assessed.

## DISCUSSION

This systematic review and meta-analysis suggests, that parents of preterm infants admitted to SFR NICUs experienced better outcomes compared with parents of infants admitted to OBU NICUs. We found lower NICU-related stress levels upon discharge, and more parental presence, involvement, skin-to-skin care, empowerment, degree of FCC, and satisfaction levels in parents of preterm infants admitted to SFRs. No differences were found in anxiety, parent-infant bonding, or depression upon or after discharge. No studies examined the association of different care environments with post-traumatic stress.

The findings of this review can only be generalised to mothers of preterm infants because only four studies included fathers[131,181–183], and only two studies examined well-being outcomes in fathers.[131,183] Fathers can feel stressed, depressed, excluded, isolated, and incompetent during and after hospitalisation of their infant in the NICU.[61] Additionally, the effect of infant hospitalisation on fathers might be different than on mothers.[188] More research that focuses on outcomes for fathers is necessary. Specifically, it is important to focus on the (different) role fathers play during hospitalisation of their infant supporting the infant and the mother, which might not be adequately captured when using questionnaires validated in mothers.

Almost all included studies showed that SFRs appear to facilitate parental presence, involvement, and SSC, supporting the WHO and EFCNI recommendations.[76–78] Engaging parents in their infants' care may lead to favourable long-term outcomes not only in infants[55,134,135,172] but also in the parents themselves.[55,189] We show this in subgroup analyses, focusing on studies with high parental presence and SSC levels in SFRs; in these studies, lower depression and stress levels were present in parents of preterm infants. This suggests a moderating effect of parental presence, SSC and involvement in the association between SFR and outcomes not only in infants but also in parents.[134,135]

Further research is required to understand the specific factors during hospitalisation of preterm infants that improve the outcomes for parents. This is especially important when hospital budgets are constrained, and priorities need to be established. We need to understand whether or not SFRs are required or if some of associated benefits can be achieved with other family-centred approaches such as family-centred rounds with supported parent participation and presence, increased support for parents to provide developmentally supportive care, better communication with parents, parent education, or family integrated care models.[55,69,70,134] Also, caregiving practices for mothers, for instance couplet-care was heterogeneous in included studies, and should be studied more in depth. Other facilities might also be beneficial for parents, for instance a kitchen, lounge room, comfortable chair or other purpose-built family accommodations.[190] Teasing out specific factors and understanding their impact requires detailed data collection from individual families during a time in which they are already under stress. Mediation analyses or network meta-analyses[191] might be able to clarify the beneficial (associated) factors in SFRs.

By including not only RCTs, but all comparative study designs, we have created a complete overview of the existing literature that is highly generalisable to the neonatal field. However, only one randomised trial (with high internal validity) was found.

Therefore, the overall quality of evidence on the effect of SFR on parental well-being is low. Many studies were considered to have serious RoB, specifically in the selection of participants, the measurement of outcomes, and confounding. This was mainly because the interventions studied were inevitably hospital-level interventions for which randomisation is difficult. Therefore, we cannot claim a causal relationship between SFRs and improved parent well-being. For instance implementing SSC might be easier in single family rooms, but could also be a result of care culture in the unit.[192] Also, statistical heterogeneity was high for many outcomes. Although this decreased in the sensitivity analyses if RoB was considered for stress, anxiety and depression, only a paucity of studies was available to explore this in-depth. More (robust) research is needed to provide more insight into the association between SFR and improved well-being in parents. In the future, (stepped wedge) cluster randomised trials should be considered to investigate hospital-level interventions, as these are less prone to bias than non-randomised trials.[55,109]



The data collection methods on parental presence, involvement, and SSC have not yet been validated and it presents an ongoing challenge to collect this information accurately without being subjective. Almost all outcomes were by necessity self-reported outcomes, which in an unblinded study are more prone to measurement bias. Biomarkers for outcomes could be a potential solution, but was only used in one study to measure for stress in parents.[112,193] Whenever using biomarkers and specifically cortisol, several confounding factors should be considered, which potentially influence the outcome. For cortisol levels in saliva, the sampling time should be taken in consideration.[194] As we used summary measures as provided by the paper, we did not know how sampling times influenced the levels of stress in the baseline measures, as this study was designed to compare stress reactivity and co-regulation between groups.

Parental stress in the NICU has been described in multiple studies and is associated with longterm health of parents and their offspring.[195] However, none of the included studies assessed parent well-being beyond four months after discharge. As the risk of psychological distress is known to persist into early childhood, longer follow-up studies are needed.[196] In addition, important contextual factors such as personality traits, pregnancy and birth experiences, and family factors should be included in future studies.[188] Although post-traumatic stress symptomatology is common in parents of preterm infants[57] it was not measured in any of the studies in this review and should be addressed in future research.

This systematic review suggests that single family rooms facilitate parental presence, involvement and skin-to-skin care, and are associated with improved outcomes in parents during preterm infant hospitalisation. Most studies were characterised by serious risk of bias. Therefore, more robust research is needed as single family rooms seem to be a promising hospital level intervention for this vulnerable patient population and their families.

**Table 1. Characteristics of studies, study populations and outcomes measured in parents of preterm infants**

| First Author [reference] | Population | Country | Study type | Start of SFR care | Children in SFR | Mothers in SFR | Fathers in SFR | GA (wks (SD) or range) in SFR | Birthweight (grams (mean (SD) or range) in SFR | Children in OBU (n) | Mothers in OBU | Fathers in OBU | GA (wks (SD) or range) in OBU | Birthweight (grams (mean (SD) or range) in OBU | Presence (P), involvement (I) or SSC(S))* | Stress at discharge | Stress after discharge | Anxiety at discharge | Anxiety after discharge | Depression at discharge | Despression after discharge | Self-efficacy, empowerment and use of resources | Satisfaction | Family-centred care | Parent-infant bonding |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Rosenblum [121] | 1 | USA | BA | AA | 102 | NR | NR | 29·4 (0·3) | 1505 (52) | 106 | NR | NR | 29·5 (0·3) | 1457 (48) | - | - | - | - | - | - | - | - | Y | - | - |
| Erdeve[114] | 2 | Turkey | NRPI | WS | 31 | 26 | 0 | 30·8 (1·7) | 1452 (82) | 29 | 23 | 0 | 30·4 (2·1) | 1413 (351) | Y (P) | - | - | - | - | - | - | Y | - | - | - |
| Erdeve[113] | 2 | Turkey | NRPI | WS | 31 | 26 | 0 | 30·8 (1·7) | 1452 (82) | 29 | 23 | 0 | 30·4 (2·1) | 1413 (351) | Y (P) | - | PSI-SF | - | - | - | EPDS | - | - | - | - |
| Mörelius[112] | 3 | Sweden | RCT | WS | 152 | 135 | 0 | 33·2 (2·8) | NR | 137 | 120 | 0 | 32·6 (2·9) | NR | Y (P/I) | Biomarker | - | - | - | - | - | - | - | - | - |
| Pineda[116] | 4 | USA | NRPI | AA | 42 | 42 | 0 | 26·81 (0·82) | NR | 39 | 39 | 0 | 26·31 (1·91) | NR | HR (P/I/S) | PSS-NICU† | - | STAI | - | EPDS | - | - | - | - | - |
| Wataker[117] | 5 | Norway | NRPI | AA | 36 | 31 | 0 | 32·7 (4·0) | 1900 (600) | 30 | 30 | 0 | 34·7 (2·2) | 2600 (838) | - | - | - | - | - | - | - | Y | - | - | - |
| Flacking[118] | 6 | Sweden | NRPI | AA | 114 | 96 | 0 | 32·7 (2·9) | 2046 (712) | 186 | 170 | 0 | 32·1 (2·8) | 1980 (657) | SR (S) | - | SPSQ | - | - | - | - | - | - | - | - |
| Lester[134] | 7 | USA | BA | AA | 252 | 235 | 0 | 28·3 (2·4) | 1050 (278) | 151 | 147 | 0 | 28·2 (2·3) | 1033 (261) | HR (P/I/S) | PSS-NICU | - | - | - | - | - | - | PG | FCCS | - |
| Lester[135] | 7 | USA | BA | AA | 123 | 123 | 0 | 26·9 (1·7) | 914 (220) | 93 | 93 | 0 | 27·1 (1·7) | 938 (248) | HR (P/I/S) | PSS-NICU | - | - | - | BDI | - | - | - | - | - |
| Blomqvist [184] | 8 | Sweden | NRPI | AA/WS | 49 | 49 | 49 | 32·1 (28·7 – 33·7) | 1760 (740-2920) | 55 | 55 | 55 | 32·1 (28·4 – 33·9) | 1870 (930 – 2625) | SR /HR (S) | - | - | - | - | - | - | - | - | - | - |

4

**Table 1. Characteristics of studies, study populations and outcomes measured in parents of preterm infants (continued)**

| First Author [reference] | Population | Country | Study type | Start of SFR care | Children in SFR | Mothers in SFR | Fathers in SFR | GA (wks (SD) or range) in SFR | Birthweight (grams (mean (SD) or range) in SFR | Children in OBU (n) | Mothers in OBU | Fathers in OBU | GA (wks (SD) or range) in OBU | Birthweight (grams (mean (SD) or range) in OBU | Presence (P), involvement (I) or SSC(S))* | Stress at discharge | Stress after discharge | Anxiety at discharge | Anxiety after discharge | Depression at discharge | Despression after discharge | Self-efficacy, empowerment and use of resources | Satisfaction | Family-centred care | Parent-infant bonding |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Baylis[185] | 8 | Sweden | NRPI | AA/ WS | 38 | 38 | 38 | NR | NR | 43 | 43 | 41 | NR | NR | SR /HR (S) | - | - | - | - | - | - | - | - | - | - |
| Jones[131] | 9 | AUS | BA | AA | 32 | 29 | 3 | 32·8 (2·7) | 1939 (691) | 49 | 46 | 3 | 32·5 (5·3) | 1899 (495) | SR (P/S) | PSS-NICU, DASS-21 | DASS-21 | DASS-21 | DASS-21 | DASS-21 | DASS-21 | PES | - | - | - |
| Raiskila[187] | 10 | Europe | NRPI | AA/ WS‡ | 108 | 108 | 107 | NR | NR | 103 | 102 | 96 | NR | NR | SR (P/I/S) | - | - | - | - | - | - | - | - | - | - |
| Aija[186] | 10 | Europe | NRPI | AA/ WS‡ | 100 | 88 | 76 | NR | NR | 72 | 68 | 54 | NR | NR | SR (P/I/S) | - | - | - | - | - | - | - | Y | Y | - |
| Tandberg[182] | 10 | Norway | NRPI | AA | 33 | 29 | 28 | 33 (1·7) | 1889 (473) | 31 | 29 | 29 | 31·1 (3·0) | 1643 (679) | SR (P/I/S) | - | - | - | - | - | - | - | - | Y | - |
| Tandberg[181,183] | 11 | Norway | NRPI | AA | 35 | 30 | 30 | 30·5 (NR) | 1452 (301) | 42 | 36 | 36 | 30·1 (NR) | 1382 (274) | SR (P/I/S) | PSS-NICU, PSI-SF | PSI-SF | STAI-SF | STAI-SF | EPDS | EPDS | - | - | - | MPAS |
| **Total** | **11** | | | | **923** | **781** | **189** | | | **927** | **768** | **190** | | | **9** | **5** | **4** | **3** | **2** | **4** | **3** | **3** | **3** | **2** | **1** |

*: Appendix Table 5, †:Parental role alteration subscale, ‡ : different in the participating units, AA: at admission, AUS: Australia BA: before-after study, BDI: Beck Depression Inventory, EPDS: Edinburgh Postnatal Depression Scale, FCCS: Family-Centred Care Survey, HR: healthcare-professional reported, I: Involvement, MPAS: Maternal Postnatal Attachment Scale, NA: not applicable, NR: not reported, NRPI: non-randomised prospective study, P: presence, PES: Parent Expectations Scale, PG: Press Ganey NICU Survey, PSI: Parenting Stress Index, RCT: randomised controlled trial, SPSQ: Swedish Parenting Stress Index, SR: self-report, S/SSC: skin-to-skin care, STAI: State-Trait Anxiety Inventory, USA: United States of America, WS: when stable, Y: yes

**Table 2. Risk of bias in included studies**

| Source | Bias due to confounding | Bias in selection of participants into the study | Bias in the classification of intervention | Bias due to deviations from intended interventions | Bias due to missing data | Bias in the measurement of outcomes | Bias in selection of reported result | Overall RoB |
|---|---|---|---|---|---|---|---|---|
| [121] | Moderate | No information | Low | Low | No information | No information | No information | No information |
| [113,114] | Serious | Serious | Low | Low | Moderate | Serious | Moderate | Serious |
| [117] | Serious | Serious | Low | Low | Serious | Serious | Moderate | Serious |
| [116] | Moderate | Serious | Low | Low | Low | Serious | Moderate | Serious |
| [118] | Serious | Serious | Low | Low | Moderate | Serious | Moderate | Serious |
| [184,185] | Serious | Serious | Moderate | Low | Low | Serious | Moderate | Serious |
| [131] | Low | Serious | Low | Low | Serious | Serious | Moderate | Serious |
| [134,135] | Low | Serious | Low | Low | Serious | Serious | Moderate | Serious |
| [187] | Critical | Critical | Moderate | Low | Serious | Serious | Moderate | Critical |
| [181–183] | Serious | Serious | Low | Low | Low | Serious | Moderate | Serious |
| **Cochrane** | **Bias arising from the randomization process** | | | **Bias due to deviations from intended interventions** | **Bias due to missing outcome data** | **Bias in the measurement of the outcome** | **Bias in selection of the reported result** | **Overall RoB** |
| [112] | Low | | | Low | Some concerns | Low | Low | Some concerns |

See Appendix for an elaborate discussion on RoB assessment

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**Table 3. Meta-analyses of single family rooms versus open bay units on outcomes in parents of preterm infants**

| Outcome | Subgroup analyses | Study populations (n) | Total parents (n) | Parents in SFR (n) | Parents in OBU (n) | Heterogeneity ($I^2$, %) | SMD | 95%CI | P-value |
|---|---|---|---|---|---|---|---|---|---|
| During hospital stay | | | | | | | | | |
| Participation | Presence | 6 [114,116,131,134,135,181,187] | 892 | 486 | 404 | 98% | - | - | - |
| | Involvement | 2 [134,182] | 497 | 292 | 205 | 78% | - | - | - |
| | SSC | 5 [116,118,134,135,183,187] | 993 | 551 | 542 | 96% | - | - | - |
| Upon discharge | | | | | | | | | |
| Stress* | *All* | 5 [112,116,131,134,135,183] | 909 | 496 | 413 | 79% | - | - | - |
| | *Biomarker* | 1 [112] | 255 | 135 | 120 | NA | -0·07 | -0·32; 0·01 | 0·56 |
| | *NICU- related~* | 3 [131,134,183] | 573 | 319 | 254 | 0% | -0·41 | -0·58; -0·24 | <0·0001 |
| | *Parenting~* | 0 | - | - | - | - | - | - | - |
| Depression | *All* | 4 [116,131,135,183] | 488 | 249 | 239 | 87% | - | - | - |
| Anxiety | *All* | 3 [116,131,183] | 272 | 126 | 146 | 81% | - | - | - |
| Follow-up (2 to 4 months after discharge) | | | | | | | | | |
| Stress | *All* | 4 [113,118,131,183] | 451 | 183 | 268 | 32% | -0·09 | -0·35; 0·17 | 0·495 |
| | *Biomarker* | NA | - | - | - | - | - | - | - |
| | *NICU- related~* | NA | - | - | - | - | - | - | - |
| | *Parenting~* | 3 [113,118,183] | 419 | 171 | 248 | 50% | -0·12 | -0·43; 0·19 | 0·466 |
| Depression | *All* | 3 [113,131,183] | 185 | 87 | 98 | 16% | -0·15 | -0·48; 0·18 | 0·372 |
| Anxiety | *All* | 2 [131,183] | 136 | 61 | 75 | 0% | -0·17 | -0·51; 0·17 | 0·316 |

*see Appendix for the different stress constructs measured, †STAI-state anxiety, 95%CI: 95% confidence interval, NA: not applicable/available, OBU: open bay unit, OR: odds ratio, SFR: single family room, SMD: standardised mean difference. See Appendix p.20 for sensitivity analyses.

Chapter 5

# An international study on implementation and facilitators and barriers for parent-infant closeness in neonatal units.

## Zero separation in neonatal care: a qualitative study

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# ABSTRACT

### Importance

Parent-infant closeness and active parent participation in neonatal care are important for parent and infant health.

### Objective

To give an overview of current neonatal settings and gain in-depth understanding on facilitators and barriers for parent-infant closeness, zero-separation, in 19 countries.

### Methods

Neonatal Intensive Care Unit (NICU) professionals, representing 45 NICUs from a range of geographic regions in Europe and Canada, were purposefully selected and interviewed June-December 2018. Thematic analysis was conducted to identify, analyze and report patterns (themes) for parent-infant closeness across the entire series of interviews.

### Results

Parent-infant separation during infant and/or maternity care is very common (42/45 units, 93%), despite implementation of family integrated care (FICare) practices, including parent participation in medical rounds (17/45, 38%), structured education sessions for parents (16/45, 36%) and structured training for health care professionals (22/45, 49%). NICU professionals encountered four main themes with facilitators and barriers for parent-infant closeness on and between the hospital, unit, staff and family-level: Culture (jointly held characteristics, values, thinking and behaviours about parental presence and participation on the unit), Collaboration (the act of working together between and within different levels), Capacities (resources and policies), and Coaching (education to acquire and transfer knowledge and skills).

### Interpretation

Implementing parent-infant closeness in the NICU is still challenging for health care professionals. Further optimization in neonatal care towards zero-separation and parent-infant closeness can be achieved by enforcing the ‘four Cs for Closeness’: Culture, Collaboration, Capacities, and Coaching.

## INTRODUCTION

Preterm and ill infants can spend considerable time in the neonatal intensive care unit (NICU) after birth before going home with their parents. This period (during infant and/or mother hospitalization) is often characterized by parent-infant separation, limiting emotional and physical closeness between parents and their infants.[31,49] In the NICU, parents sometimes feel they cannot take on typical parenting roles. Parents can suffer from feelings of helplessness, they can experience high levels of stress or trauma, they can feel unprepared to go home, which all can potentially impact parent and infant health.[49,51,59,197]

Minimizing parent-infant separation (for example through parent-led interventions such as skin-to-skin care (SSC)[66] or the implementation of couplet-care[67]) as well as endorsing parent-partnership within the infant's care team, and involvement and integration in neonatal care are associated with health benefits for infants and their parents[34,55,59,68–74] and advocated by parent representatives and the World Health Organizatio.[75–78] Parent-infant closeness and zero-separation has received particular attention over the past years alongside the increasing interest and implementation of family integrated care (FICare).[49,63,93] FICare uses a comprehensive framework that endorses parent-provider partnership and parent-infant interaction by enabling parents as primary caregivers in the neonatal unit and as equal partners in the care team.[55,93] Parent-infant closeness is a core component and outcome of FICare[93] as parents can experience a sense of closeness during NICU care when enacting parental roles, especially autonomously and when making decisions concerning their infant.[51,198] Nevertheless, parents can still experience less empowerment, stress and separation from their newborn when co-care for the mother-infant dyad is not provided.[118,199,200]

Little is known on the current state and application of parent-infant closeness in European NICUs. Above, it is unclear which barriers health care professionals encounter to keep families close and enable them to participate in neonatal care during NICU stay.[70] Previous studies have mainly described access policies for families in the NICU, availability of single bed units, compliance with the Baby-Friendly Hospital Initiative, and actual parental presence, SSC or participation in medical rounds.[186,187,201–204] Other work has focused on the concept, pathways and feelings of closeness from parents' perspectives[51,198,205], insights into perceptions and aspirations of highly motivated medical staff to physical closeness[192] and facilitators and barriers for family centered care from staff employed in hospitals from three European countries.[206] To our knowledge, no data or qualitative analysis is currently available considering facilitators to implement parent-infant closeness and to achieve zero-separation in neonatal care covering a vast majority of European countries.

The objective of this study was therefore to give a comprehensive overview and gain in-depth understanding of current neonatal settings and facilitators and barriers with regard to parent-infant closeness during infant hospitalization in 19 countries.

# METHODS

It is difficult to understand neonatal care practices (specifically mother-infant care) and to interpret care models from survey data in the absence of validated questionnaires and clear definitions. Therefore, we used a qualitative study design, conducting in-depth interviews with NICU professionals. The data collection tool was a semi-structured interview guide developed in collaboration with parents (see Supplement). *Parent-infant closeness* (zero-separation) was defined as "the possibility to be together (emotionally and physically) under all (medical) circumstances and according to parents' preferences, approximating the situation in full term infants and the family is home." *Parent participation in neonatal care* was defined as previously.[207]

## Participants

We aimed to include a geographically and culturally diverse sample with ≥2 hospitals per European country. Health care professionals (mainly pediatricians/neonatologists) of NICUs, able to provide care to infants <30 weeks of gestation, were contacted by e-mail through the international network of one of the authors (JBvG, former board member of the European Society of Pediatric Research (ESPR) and the European Society of Pediatric Gastroenterology, Hepatology and Nutrition (ESPGHAN)). This resulted in participants from 11 countries. We used a purposive sampling methodology with additional snowballing to either include experts in parent-partnered neonatal care (PPNC) models[70] or to contact units in neighbouring countries. Examples of experts included were the Close Collaboration with Parents research group[54] and hospitals in Sweden and Denmark known for their PPNC practices. We included the Canadian site as they have a unique pioneering role concerning worldwide implementation and dissemination of FICare[55], and therefore their view was indispensable for our research question. Even though they were not situated in Europe, we decided to include them for richness of data. PPNC experts were units with peer-reviewed publications on PPNC models and/or units that trained other sites on PPNC.

## Data collection

With participants' permissions, interviews were audio- and video-recorded between June and December 2018. Interviewees were invited to elaborate on answers, and follow-up and probing questions were asked when limitations and possibilities for parent-infant closeness were encountered. The interviews lasted approximately 30 to 60 minutes and were conducted by one interviewer and were video-recorded with Zoom(Zoom Video Communications, San Jose, CA, USA). The interviewer had no prior relationship to the interviewees. All interviews were conducted in English, except for two interviews with professionals from Ukraine, for which a person fluent in Russian and Ukrainian translated during interviews (see Acknowledgments).

## Data analysis

Thematic analysis was used to identify, analyze and report patterns (themes) across the entire series of interviews. Data analysis started after all interviews were transcribed verbatim and transcripts were returned for comment and/or correction from participants. Data analysis was performed by NRvV and NHML and reviewed with a parent representative (SM). For details on the research team, see the Supplement.



We followed the six-steps as outlined by Braun and Clarke[208] and used MAXQDA 2007[209] with a hierarchical coding structure to code interviews. Two investigators iteratively developed the coding scheme. We used a combined inductive and deductive approach. Sensitising topics for the deductive approach were logistics and architecture of the unit, parent participation, education of parents, and education of staff based on previous literature and clinical experience of the multidisciplinary team. Additionally, we coded with new codes if new facilitators and barriers were encountered in the interviews (inductive approach). To avoid interpretative bias, first 3 interviews were independently coded and then discussed. Following, another 3 interviews were independently coded to refine the codebook, and again discussed to resolve discrepancies. After, NRvV and NHML discussed with the primary author group on the final codebook. One author (NRvV) coded all remaining interviews. We believe data saturation was achieved as no new codes arose. Subsequently, NRvV and NHML discussed all codes and grouped them into themes and discussed the relation and inter-relatedness between themes within an iterative process. Identified themes were reviewed by the research team to help contextualize and reorganize the themes from a multidisciplinary perspective.

## Ethics approval and use of checklists

We used the Consolidated criteria for reporting qualitative research(COREQ)-checklist for interviews and focus groups[210] and the Guidance for Reporting Involvement of

Patients and the Public short form(GRIPP2) reporting checklist.[211] Ethical permission to undertake the study was given by the Institutional review board of Amsterdam UMC, location AMC, the Netherlands.

## RESULTS

Health care professionals from 46 hospitals were asked to participate. Forty-five (98%) consented to collaborate, representing 19 countries (18 countries were situated in Europe, Figure 1, Table 1, Supplement) and mainly medical doctors working on the neonatal unit (91%).

**Figure 1. European countries participating in the Creating Room and Opportunities On Wards for Newborns and their families (CROWN) study.**

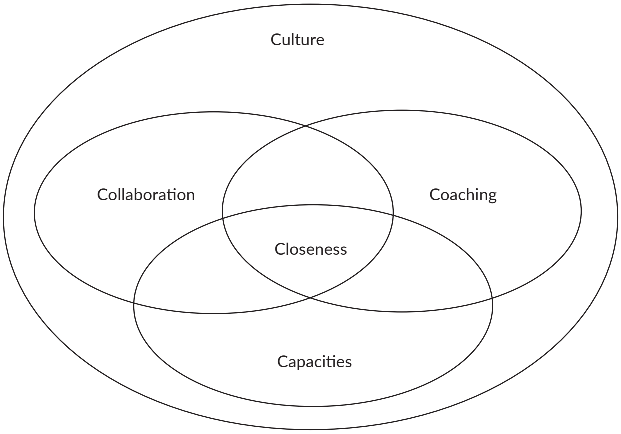
**Table 1. Characteristics of participating NICUs**

| Characteristic | N = 45 |
|---|---|
| European NICU (n (%)) | 44 (98%) |
| Expert in parent-partnered neonatal care (n (%)) | 6 (13%) |
| Pediatrician/neonatologist interviewed | 41 (91%) |
| Number of beds in unit (median, IQR) | 21 (15 – 37) |
| Number of births in facility (median, IQR) | 3,400 (2,500 – 6,000) |
| Number of admission to NICU per year (median, IQR) | 500 (285 – 1100) |
| Number of VLBW (<1500 grams) per year (median, IQR) | 100 (55 – 145) |
| Level 3 NICU (n (%)) | 42 (93%) |
| Able to provide ECMO (n (%)) | 7 (16%) |
| Open access policy (n (%)) | 32 (71%) |
| Possibility to breast pump on the ward (n (%)) | 44 (98%) |
| Reclining chair available next to infant (n (%)) | 42 (93%) |
| Webcam available to see infant from home (n (%)) | 3 (7%) |
| Rooming-in possible before discharge to home (n (%)) | 41 (91%) |
| Single family room plan | |
| Yes, for all patients (n (%)) | 4 (9%) |
| For specific patient populations (n (%)) | 16 (36%) |
| No (n (%)) | 25 (55%) |
| Facility near hospital where parents can stay (n (%)) | 31 (69%) |
| Early discharge program (n (%)) | 9 (20%) |
| Parent participation in medical rounds (n (%))† | 17 (38%) |
| Structured education sessions for parents (n (%))† | 16 (36%) |
| Structured training for health care professionals (n (%))† | 22 (49%) |
| Mother-infant separation during infant or maternal care (n (%)) | 42 (93%) |

†Components of FICare, ECMO: extra corporeal membrane oxygenation, IQR: interquartile range, n: number, NICU: neonatal intensive care unit, VLBW: very low birthweight

Despite implementation of (components of) FICare, parent- and specifically mother-infant separation during maternity and/or neonatal care was very common (93%). We identified four themes for facilitators and barriers around parent-infant closeness on the hospital, unit, staff, and family-level: *Culture*, *Collaboration*, *Capacities*, and *Coaching* (Figure 2, Table 2). *Culture* was the overarching theme in our analyses, encompassing the other themes. Examples of facilitators are depicted in the Supplement.

**Figure 2. Themes concerning facilitators and barriers for parent-infant closeness in neonatal care**




### Culture

*Culture* was described as jointly held characteristics, values, thinking, and behaviors of people in workplaces and organizations. For example, at the organizational (hospital- and unit-)level PPNC expert hospitals regarded parents in hospital or unit boards to be important. For the workplace (unit- or staff-)level, culture included professionals' attitudes towards parental presence and participation in care.

Parents' participation in infant care and continuous parental presence were facilitated if hospital- or unit-management and staff had open mind-sets: "*The staff knows, we try to involve parents. But it's hard for the staff to change their mind about it. Step-by-step the department and staff are becoming more open for parents.*" [unit 25].

The workplace *Culture* varied from very natural: "*It's within the culture of the unit, the nurses are usually really upset if the parents are not there. They come to the doctor and complain [...] there are alarm clocks ringing if the parents are not there*" [unit 37] to reluctant: *"I think staff don't want parents around, because parents look on their hands"* [unit 4]. Also, parents could feel unwelcome on the unit due to negative professional attitudes: "*Sometimes there can be a nurse, that is not so nice, and parents feel not good unfortunately. It is the attitude of everyone how welcome you feel*" [unit 4].

A *Culture* change was needed on the staff level, influencing current beliefs of power and hierarchical structures between professionals and parents to promote parent-infant closeness: "*It's so easy for the medical staff to dictate parents what to do, you keep the same* [hierarchical] *power structure. It's hard for us to give the power to the family* *and ask them what they want to do and create a welcoming atmosphere that they want to stay there"* [unit 36].

**Collaboration**

*Collaboration* is working together on and between the different levels. The current (historical) division and compartmentalization between the maternity and neonatal departments within the hospital limited parent- and specifically mother-infant closeness in the postpartum period. Poor *Collaboration* and co-organization on the hospital, unit and staff level impeded parent-infant closeness: "*We always fight with them [*about gavage feeding in the maternity ward], *but we are different organizations so we are not the same"* [unit 13].

Staff from different units and disciplines had to collaborate and work together to keep families close: "*Sometimes we have an agreement with the maternity ward that they come and supervise us and take care of the mother and they can stay with their child"* [unit 35]. In one unit, midwives were on the NICU department's payroll and always present in the NICU for maternity care, others had full obstetric and neonatal co-organization of unit, care and staff.



Parent-staff *Collaboration* was also important. Some professionals referred to a distinct and added role for parents, enhancing *Collaboration* with the healthcare team: "*It is kind of promoted that everybody has their own important role to play. Parents carry that sixth sense"* [unit 12] and during painful procedures: *"A very important role for parents is pain management, non-pharmacological [...] when they have this role to comfort the baby, they hold the baby, and many times we do not need pain medication"* [unit 36].

Family centred rounds (FCRs, 17/45, 38%) were important for parent-staff *Collaboration*. However, organizing schedules between parents and professionals could be challenging. Important were professional beliefs on the added and distinct role of parents: "*I think FCRs are very wise. The parents could give us a lot of information about the infant"* [unit 4]. Privacy issues limited FCR implementation in NICUs with open bay settings: "*Parents cannot be in the unit because of confidentiality questions"* [unit 3]. Some NICUs provided solutions in these settings such as: headphones for other parents during FCRs and signing confidentiality agreements. Some professionals regarded FCRs less efficient: "*Parents used to be present during daily rounds. But now we changed the way that we are doing rounds. It is in a separate room, so they are not invited anymore. It is more efficient and faster"* [unit 41]. Other interviewees described increased efficiency, as parents often do not require extra information after the FCR. In single family rooms (SFRs), privacy was not an issue, and SFRs contributed to parent-staff *Collaboration*.

**Capacities (resources and policies)**

*Capacities* included resources of supply or support which could be physical (e.g., equipment) or human (e.g., staffing). Policies included guidelines that determined courses of action (e.g., welcoming policies) as well as logistics.

***Resources***

On the hospital level, obstetric and maternity departments were sometimes distant from the NICU, preventing mothers from being with their baby continuously: "*Our maternity ward is in another building*" [unit 16].

At the unit level, SFRs (available in 20/45, 44%) were facilitators for parent physical presence at the bedside of the infant, but also other amenities (independent of SFRs) were important for 24-hour presence such as a bed, a kitchen, a bathroom, and comfortable chairs. Lack of space limited closeness between parents and their infant: "*The big difficulty for us is space, you know, we've got several babies in a room with lots of equipment, and it is very difficult*" [unit 3].

For staff, available time, (under)staffing issues, and high patient loads hindered parent-infant closeness: "*We don't like to give an intravenous catheter on the maternity ward, as it is a very busy ward and if these nurses need to take care of these babies then they do not take care of other babies–they do not have time to do everything*" [unit 24].

Resources (such as maternity beds and midwives in the NICU and gavage feeding or phototherapy in maternity wards) impacted parent-infant closeness. More than half of the hospitals had patient hotels or Ronald McDonald Houses® (31/45, 69%), where parents could stay after mother was discharged (usually 5-7 days after birth). However, sometimes these were earmarked for parents living far away and not always available to all parents. Many NICUs indicated rooming-in before discharge to home to be important and to be common practice (41/45, 91%).

Several family resources were important for family presence, including financial resources, distance to hospital, and family composition. Professionals were ambivalent about the implementation of IT-applications and specifically a webcam to achieve closeness between parents and their infants during hospital stay (available in 3/45, 7%): "*No, we want them* [the parents] *to be present in the unit*" [unit 35].

***Policies***

On the hospital level, free meals and free parking for parents were deemed essential and on the unit level, policies for parent-infant closeness included promotion of (early)

SSC, promotion of breastfeeding and availability of breastpumps (44/45, 98%), parent participation in care and decision-making, and open access policies (32/45, 71%). Mother- and infant health determined logistics: "*Babies can go there* [mother-baby-unit] *as long as they are showing signs that they can feed on their own*" [unit 3]. Often logistics limited parent-infant closeness, because mothers would usually be transferred consecutively from antenatal wards to delivery rooms, to maternity wards, and then to home, which was different from the infant hospital stay in the NICU.

Early discharge and homecare programs (9/45, 20%) facilitating gavage-feeding and cardio-respiratory monitoring for families at home were important, but lack of structured education for parents and sometimes far distances inhibited successful implementation.

**Coaching**

Finally, *Coaching*, often referred to as "education", acquiring and transferring knowledge and skills, was important. Currently, a discrepancy between professionals' training and specifically what the reality of keeping mothers and infants together postnatally requires them to do, is present. If staff was not educated and did not feel comfortable in taking care of either mothers (NICU-professionals) or infants (obstetric or maternity care professionals), parent-infant closeness was limited: "*Maternity nurses and midwives are not comfortable with providing that sort of care*" [unit 39]. Some units exchanged or collaborated between staff: "*We have had this rotation; our NICU nurses went to the prenatal and postnatal ward and midwives were in our unit for periods*" [unit 36]. Coaching staff was either implemented regularly by structured training programs in the unit (22/45, 49%), at start of working in the unit by senior staff, or by parents. *Coaching* was enhanced by *Culture* and *Collaboration*: "*You need to provide education for staff because they support families involved. That is a big part of what happens. When a new nurse starts in the unit they are orientated in integrated care and coached by a parent*" [unit 40].

*Coaching* at the family-level was important, for families to acquire knowledge on the special care and needs of their infant and to promote parent-infant interaction. Education sessions were implemented structurally in 16/45 (36%) units.

## Relationships between themes

Culture was the overarching theme in our analyses, encompassing the other themes, centred around Closeness (Figure 2). A Culture can be nurturing for Closeness through Coaching, Collaboration, and Capacities. Collaboration and a Culture of Collaboration to achieve Closeness was characterized by co-working and coordination of care between same levels (e.g. the neonatal and obstetric units working with each other) and

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between different levels within the hospital (e.g. Collaboration between staff and the unit management, and Collaboration between parents and staff). Collaboration could be limited by Capacities (e.g. staffing issues) but facilitated by Coaching (e.g. Coaching neonatal nurses on maternity care). Coaching related to Culture (some hospitals find teaching very important with many professionals in training), but also to Capacities (e.g. dedicated professionals organizing educational sessions) and Collaboration (educated on importance of Collaboration and Collaboration between professionals facilitated Coaching).

# DISCUSSION

In this study we describe current practices in neonatal care with facilitators and barriers for parent-infant closeness in 19 countries. Despite willingness to facilitate parent-infant closeness, many barriers exist that prevent zero-separation in a vast majority of the units examined.

NICU professionals encountered challenges for parent-infant closeness within four main themes of facilitators and barriers on and between the hospital, unit, staff, and family-level: *Culture* (jointly held characteristics, values, thinking and behaviours), *Collaboration* (the act of working together between and within different levels), *Capacities* (resources and policies), and *Coaching* (education to acquire and transfer knowledge and skills).

This study provides tangible and comprehensive data to support the call-to-action to achieve zero-separation during neonatal care[63,67], and shows where priorities should be given.

Enabling parents' participation in care and presence can give them a sense of closeness.[51] However, implementing the four pillars of FICare alone might not be enough to facilitate parent-infant closeness and zero-separation.[55] We show, that despite implementation of FICare components and knowledge on the negative health effects of parent-infant separation[31,212] health care professionals still encounter challenges to keep families close. Especially, keeping mothers and infants together during specialized neonatal or maternity care is no common practice yet, and attention should be given to this topic in future innovations.[31,67,200,213]

Change within hospitals and units can be very challenging, specifically when it concerns hospital(care) culture[214], which we found was the overarching theme within our analyses in concordance with previous research.[93,206,215] The SFR design (as part of *Capacities*) has long be promoted to be the solution, but this might solely increase parental *presence* and not necessarily parental feelings of closeness or participation in care.[68,216] Above, without *Coaching* or *Collaboration* the SFR-design alone may not be the solution. And therefore, endorsing facilitators within the other themes could also have a potentially great impact on parent-infant closeness if budgets are under constraint.

We included the Canadian site because of their unique pioneering role concerning FICare worldwide. This might have introduced bias in sites, but due to the qualitative nature of our study and as we believe saturation was met because no new items arose during our interviews, we think these results give a fair example of the facilitators and barriers health care professionals encounter within the NICU context of included countries. Future research could purposefully search for units in other and more countries or for instance in developing countries and compare their results with ours, as FICare practices are on the rise in other parts of the world too.[217]



Interviews were held just before the emergence of the COVID-19 pandemic, which imposed even stricter policies on parental presence and participation in neonatal units, increasing parent-infant separation.[63,218,219] We show, that also before the pandemic, issues regarding parent-infant closeness were already present. The themes we encountered could therefore possibly be even more urgent and relevant as family supportive post-pandemic neonatal care practices are (re)established.

The results represent NICU professional views and we did not include obstetricians, midwives, and parents that *experienced* the healthcare in the included hospitals. Keeping families close could be different from their point of view in that same setting, and should be explored in depth in future research. Especially, we were unable to explore the perception of emotional closeness and the pathways towards emotional closeness that might be facilitated in the included units from parents' perspectives.[205] Also, the exact roles parents play in the infant's care team and their potential added role within multidisciplinary teams remains to be elucidated.

One of the strengths of our study, is that we included parents during all phases of conduct and analysis, making the results and challenges that are met meaningful for all stakeholders. Moreover, we have interviewed a large sample throughout Europe, whereas previous studies have focused either on health care professionals with high incentives for PPNC models[192,206], or on quantitative outcomes[187,192,201,202] without addressing in-depth understanding on the matter of parent-infant separation specifically.

Future research should focus on the different (aspects of) care concepts, themes and workplace cultures we encountered in our study, the fidelity of care models and the potential pathways towards outcomes of parents and their infants with for instance network meta-analyses[220,221] or mediation analyses.[73,222] As many data is arising on the benefits of parent participation in care and zero-separation[55,69,73,74,222], next studies should work with methods from an implementation science point of view to promote the systematic uptake of these clinical research findings into routine neonatal care.[223] Additionally, research should focus on an exact definition and measurement of "zero separation" in this context, as one can still feel emotionally connected without being physically present. Lastly, core outcome sets for family care are needed to be able to perform these and future robust studies.[224]

## CONCLUSION

In this study we describe current practices in neonatal care with facilitators and barriers for parent-infant closeness in 19 countries. Parent-infant separation during infant and/or maternity care is still very common in participating units.

Further optimization in neonatal care towards zero-separation and parent-infant closeness can be achieved by enforcing on the family-, staff-, unit- and hospital-level the 'four Cs for Closeness': *Culture*, *Collaboration*, *Capacities*, and *Coaching*.

**Table 2. Facilitators and examples to promote parent-infant closeness**

| Facilitators (themes) | Level | Examples |
|---|---|---|
| Culture | Family | The family is willing to go home with extra medical care |
| | Staff | The staff is open to change<br>The staff recognize that parents have knowledge on their child<br>The staff endorse that parents have their own distinguished and added role within the NICU / care of their infant<br>The staff value that parents are capable of taking care of their infant<br>The staff respect and acknowledge parents in their own choices<br>The staff feels responsible for other part of the dyad |
| | Unit | The unit welcomes parents at all times |
| | Hospital | The hospital (management) is open to change and values their employees and patients |
| Collaboration | Family | The family is able to arrange schedules and tasks between them and staff |
| | Staff | The staff can arrange schedules and tasks between them and the family<br>The staff supports the family to achieve closeness according to their personal needs, preferences and pace<br>The staff invites parents to family centred rounds<br>The staff from different departments and specialties work together to minimize separation |
| | Unit | Different specialty units are open to each other or merged with each other |
| | Hospital | The hospital (board) works together with unit, staff, and families |
| Capacities | Family | The family has resources to come to hospital |
| | Staff | The staff perceives an acceptable workload |
| | Unit | The units have resources and equipment present for the dyad<br>The unit has an open access policy<br>The unit has enough staff / acceptable patient load<br>The unit has a dedicated person to support parents and/or staff<br>The unit supports the use of IT/webcam<br>The unit is set up with single family rooms or has rooming-in rooms<br>The unit provides facilities for parents to stay 24 hours per day<br>The unit has a policy of early discharge programs<br>The unit promotes skin-to-skin care<br>The unit has a policy of rooming-in before discharge<br>The unit promotes breastfeeding |

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**Table 2. Facilitators and examples to promote parent-infant closeness (continued)**

| Facilitators (themes) | Level | Examples |
|---|---|---|
| | | The unit has breastpumps available for all mothers |
| | Hospital | The hospital supplies patients/families with free meals and parking |
| | | The hospital has a facility close to hospital where parents can stay |
| | | The hospital arranges the localization of units within hospital conveniently to support parent-infant closeness |
| Coaching | Family | The family educates staff |
| | | The family receives education on special care and needs of the infant |
| | | Veteran parents support parents during hospital stay |
| | Staff | The staff is educated on the importance of preventing parent-infant separation, family participation in care and parental presence in the unit |
| | | The staff is trained to perform care for other part of the dyad |
| | | The staff is comfortable working inter-, cross-, and transdisciplinary |
| | Unit | The unit is educated on the importance of preventing parent-infant separation, family participation in care, and parental presence in the unit |
| | Hospital | The hospital has a policy of professionals to be trained and educated regularly |

NICU: neonatal intensive care unit

Part II

# Collaboration, participation and communication

Chapter 6

# Development and psychometric evaluation of the CO-PARTNER tool for collaboration and parent participation in neonatal care

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# ABSTRACT

### Background

Active parent participation in neonatal care and collaboration between parents and professionals during infant hospitalization in the neonatal intensive care unit (NICU) is beneficial for infants and their parents. A tool is needed to support parents and to study the effects and implementation of parent-partnered models of neonatal care.

### Methods

We developed and psychometrically evaluated a tool measuring active parent participation and collaboration in neonatal care within six domains: *Daily Care*, *Medical Care*, *Acquiring Information*, *Parent Advocacy*, *Time Spent with Infant and Closeness* and *Comforting the Infant*. Items were generated in focus group discussions and in-depth interviews with professionals and parents. The tool was completed at NICU-discharge by 306 parents (174 mothers and 132 fathers) of preterm infants. Subsequently, we studied structural validity with confirmatory factor analysis (CFA), construct validity, using the Average Variance Extracted and Heterotrait-Monotrait ratio of correlations, and hypothesis testing with correlations and univariate linear regression. For internal consistency we calculated composite reliability (CR). We performed multiple imputations by chained equations for missing data.

### Results

A 31 item tool for parent participation and collaboration in neonatal care was developed. CFA revealed high factor loadings of items within each domain. Internal consistency was 0.558 to 0.938. Convergent validity and discriminant validity were strong. Higher scores correlated with less parent depressive symptoms ($r=-0.141$, 95%CI -0.240; -0.029, $p=0.0141$), less impaired parent-infant bonding ($r=-0.196$, 95%CI -0.302; -0.056, $p<0.0001$), higher parent self-efficacy ($r=0.228$, 95%CI 0.117; 0.332, $p<0.0001$), and higher parent satisfaction ($r=0.197$, 95%CI 0.090; 0.308, $p=0.001$). Parents in a family integrated care model had higher scores than in standard care (beta 6.020, 95%CI 4.144; 7.895, $p<0.0001$) and mothers scored higher than fathers (beta 2.103,95%CI 0.084; 4.121, $p=0.041$).

### Conclusion

The CO-PARTNER tool explicitly measures parents' participation and collaboration with professionals in neonatal care incorporating their unique roles in care provision, leadership, and connection to their infant. The tool consists of 31 items within six domains with good face, content, construct and structural validity.

## INTRODUCTION

Active parent participation in neonatal care during infant hospitalization in the neonatal intensive care unit (NICU) can ameliorate adverse outcomes for infants and their parents.[53,55,68,70,147,197,225] Through parent participation in neonatal care, parents can be a central part of the NICU care team, gain confidence in taking care of their infant, and prepare themselves for discharge.[4,226] Although the NICU has been incorporating parent involvement practices for decades, attention directed toward parent-partnered models fidelity and implementation through the examination of active parent participation and integration into care teams is currently lacking.[70]

Several tools have been developed and used to assess parent participation in the pediatric care setting.[227–229] In the neonatal setting, studies have mainly focused on constructs related to parent participation[230] such as the passive construct of (time) being present in the NICU or holding the infant[116,186,187,231], and health care professional recordings of parent competencies and activities.[116,134] Other tools have focused on aspects such as feeling guided or supported by health care professionals[232] or constructs related to maternal knowledge, confidence, expectations and social support within infant care engagement and risk evaluation.[230,233–235]

However, all aforementioned tools lack the assessment of parent active participation, and the inherent collaborative partnerships and processes that are currently changing the NICU environment from healthcare-led to parent-led infant care.[70] Most tools have also not included fathers from initial development. It is important to have validated tools to measure levels of parent participation and collaboration in the NICU to tailor care practices in real-time, to be able to assess parent-partnered care models such as family integrated care (FICare).[55,70] Above all, a broader measure is needed, that is not only centred around risk-evaluation but can also be used in a strengths-based approach to promote parent active participation in care and achieve better outcomes for infants and their parents.

In this study we developed and psychometrically evaluated the CO-PARTNER tool measuring parent participation and inherent collaboration with health care professionals in neonatal care during NICU hospitalization.

# METHODS

This psychometric study was conducted before and during a multicentre non-randomized prospective study on the effects of FICare on infants and their parents in a NICU level 2 context in the Netherlands[236], including a group of parents and infants who experienced family integrated care (FICare) in single family room units and a group who experienced standard care in open bay units (the AMICA study, see S1. Appendix for details on FICare and standard care in the different participating units). In the AMICA study, preterm infants admitted for at least 7 days to one of the participating wards and their parents were included. The primary outcome in the AMICA study was the effect of FICare in single family rooms on neurodevelopment of preterm infants. In the AMICA study, outcomes in parents (mothers and fathers separately) were also included as secondary outcomes in the short and longer term.[236] We excluded families if mothers or fathers had severe psychosocial problems (for instance acute psychiatric illness or if a family was under supervision of social services etc.), if death of a sibling occurred or if a congenital or metabolic syndrome was present in the infant.

Before conduct of the AMICA study, we considered parent active participation as a possible mediator in the pathway between the FICare-setting and improved health outcomes (for mothers, fathers and infants). However, as no validated measure of parent participation existed, we decided to conduct the generation, validation and psychometric evaluation of the CO-PARTNER tool before and during the AMICA study. We first included parents and health care professionals in the item generation phase using purposive sampling in May 2016-April 2017. For the validation and psychometric evaluation, we included parents who participated in the AMICA study and who filled out the CO-PARTNER tool at hospital discharge of their infant. Recruitment of the AMICA study took place May 2017-January 2020. The medical ethical review board of MEC-U in Nieuwegein, The Netherlands, approved the study and all parents provided written informed consent. The work described has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans. The AMICA trial was registered on the 23rd of December 2016 in the Netherlands Trial Registry NL6175.[236]

We used the quality checklist developed for the reporting of health-related-patient reported outcomes[237] for this study. The primary outcomes for this study were content validity, structural validity, and construct validity of the CO-PARTNER tool.

## Description of the construct to be measured

We adapted the definition as proposed by Power and Franck for parent participation, including the unique roles parents have during infant NICU stay and the process of collaboration with staff for developing capacity to perform activities independently.[238] Parent participation is defined as "The activities performed by a parent/guardian for their infant in the hospital setting in which they share, take part or independently act in the care of their infant across the entire hospital episode. Activities are defined as physical, psychological, or social performed by parents to improve the health and/or psychological well-being of their infant, with or without collaboration with health care professionals." We developed a formative measure to the concept of parent participation.

## Content validity

The Index of Parent Participation (IPP, developed for paediatric care)[228] questionnaire was used as a starting point as many of the 36 items could be completed by parents during infant hospitalization in the NICU.

### Item Generation



Two researchers (NvV and SvdS) independently and blind from each other extracted relevant items from the IPP[228] for the NICU setting. We simultaneously consulted the original author of the IPP on which items of the 36 in the original IPP could be applied to a NICU care context. This resulted in 26 items to be included in the item generation phase. Focus groups, one-on-one interviews and scoring of the instrument was performed with a purposive sample of six health care professionals and forty-five parents. Health care professionals included a speech therapist experienced in FICare and nurses and midwives who either worked at the FICare or the standard care unit, with a large range in working experience (8 to 30 years in profession). Parents (mothers or fathers >18 years of age) had a preterm infant (born at a gestational age between 24 weeks - 36 6/7 weeks), were at the time experiencing or had experienced a NICU stay in the previous 2 years, and had experience in either a standard or FICare unit participating in the AMICA trial. Parents and professionals were approached by independent researchers. Specifically for parents, the researchers were not involved in the care of their infants. Participants were asked to identify (additional) items on parent participation. Above, we investigated their views on content of items, how response options to items should be presented and on the rightful inclusion of the 26 items from the original IPP in the first version of the tool.[239] Participants were asked to score items (during generation from the original IPP, focus groups or one-on-one interviews) as; (1) relevant or not relevant in light of parent participation in the NICU; (2) if the items needed a yes/no response, or if the items had to be scored on a scale and were intended to examine a collaborative process in care towards being able to perform activities independently ('the nurse does this', 'the nurse and I do this together' and 'I do this independently'). Inclusion of participants ended after no new items were identified and consensus was reached on item responses.

The research team, health care professionals and parent consultants identified a total of 88 relevant items that could be considered meaningful to the concept of parent participation and the process of collaboration in the NICU context. Two neonatologists, a researcher specialized in parent empowerment, and one neonatal nurse, independently and blind from each other, scored the items as to their applicability to the concept of parent participation and collaboration in the NICU. If at least 3 out of 4 experts rated the item as relevant, it was included in the CO-PARTNER tool. A total number of 34 items were generated during the item generation phase but three items were dropped during the analysis phase (see Structural validity) resulting in a total of 31 items included.

### Conceptualizing Six Domains

After item generation research members consulted together on concept use, and current state in the literature.[68,70,200] Language considerations are described in the S2. Appendix. The research team identified the definition of parent participation to be multidimensional and items were applied to each domain based on informal consensus in an empirical and iterative process.

The six domains are based upon essential parent participation, collaboration and role within the NICU context: (1) *Daily Care*; (2) *Medical Care*; (3) *Acquiring Information*; (4) *Parent Advocacy*; (5) *Time Spent with Infant*; and (6) *Closeness and Comforting the Infant* (See Table 1).

**Table 1: CO-PARTNER tool**

| Activity | Response |
|---|---|
| **Domain 1. Daily Care** | |
| 1. Bath my child/clean my child with a washcloth. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 2. Change my child's diaper. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |

**Table 1: CO-PARTNER tool (continued)**

| Activity | Response |
|---|---|
| 3. Feed my child (breast or bottle). | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 4. Change my child's clothing. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 5. Get my child out of the incubator/cradle. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 6. Give my child medication. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 7. Weigh my child. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 8. Keep track of output (urination and defecation) of my child | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 9. Measure the temperature of my child. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 10. Keep track of my child's weight. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 11. Keep track of drinking and my child's feeds. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| **Domain 2. Medical Care** | |
| 12. Give tube feeding to my child. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 13. Look at my child's monitor and handling accordingly (e.g. stimulating during a bradycardia). | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |

**Table 1: CO-PARTNER tool (continued)**

| Activity | Response |
|---|---|
| 14. Regulate the visiting of others to my child. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 15. Participate in the daily rounds with the doctor. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| **Domain 3. Acquiring Information** | |
| 16. Did you ask health care professionals information on the health of your child? | o Yes<br>o No |
| 17. Did you ask the health care professionals for information about your child for times when you were not present? | o Yes<br>o No |
| 18. Did you talk with another parent about your experiences? | o Yes<br>o No |
| **Domain 4. Parent Advocacy** | |
| 19. I stood up for my child; I told somebody to do something in the care of my child. | o Yes<br>o No |
| 20. I stood up for my child; I told somebody NOT to do something in the care of my child; I gave boundaries | o Yes<br>o No |
| 21. I gave an explanation on the daily routines of my child to a health care professional. | o Yes<br>o No |
| **Domain 5. Time Spent with Infant** | |
| 22. On average, how many hours were you present in the hospital with your child? | Number of hours per day: |
| 23. On average, how many hours a day do you have contact with your child? | Number of hours per day: |
| 24. On average, how many hours were you really close with your child? | Number of hours per day: |

**Table 1: CO-PARTNER tool (continued)**

| Activity | Response |
|---|---|
| **Domain 6. Closeness and Comforting the Infant** | |
| 25. Hold/rock/cuddle my child. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 26. Comfort my child whenever he/she needs it. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 27. Kangaroo care / skin to skin contact. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 28. Be together with my child, be close with my child (intimate time). | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 29. Be together with my child (be present). | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 30. Soothe my child during a painful procedure (for instance drawing blood). | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |
| 31. Recognize my child's signals. | o The nurse does this<br>o I do this together with the nurse<br>o I do this independently (without the help of the nurse)<br>o This is not applicable |

Domains 1 and 2 consist of 11 and 4 items, respectively, and measure the nature of parent participation in activities of daily care and medical care. The degree of collaboration between parents and health care professionals is indicated by the response options. These items are measured on a 3-point scale (e.g. I do this myself/ independently; I do this together with the nurse; or The nurse does this) or scored as "This was not applicable". The following three items measure *Acquiring Information* and the next three items measure the nature of *Parent Advocacy* activities while caring for their child in the NICU. Questions are answered either yes or no. Three questions pertain to the amount of *Time Spent with Infant* in the NICU. This domain represents the mean time over the hospital stay that parents reported to be present and felt close with their child per day in hours. Seven items pertain to *Closeness and Comforting the Infant*, and include activities such as comforting the infant during painful procedures and kangaroo care, and the process of collaboration with staff is visible through the response options.

## Data collection

The tool was evaluated by fathers and mothers of infants enrolled in the AMICA study, a prospective non-randomized study evaluating the effect of a family integrated care model in level 2 NICUs in the Netherlands (see S3. Appendix for an elaborate description of the neonatal population and caregiving practices in the Netherlands). Questionnaires were sent using Castor Electronic Data Capturing[240] at admission and at discharge from the level 2 NICU. In the case of families with multiple births, fathers and mothers received 1 questionnaire per time point. Parents received 2 reminders if they did not fill out the questionnaire (1 and 2 weeks after the initial questionnaire was sent). All parents completed a survey package that included the tool and additionally, surveys on perceived stress in the NICU (PSS-NICU)[241], depression and anxiety[242] (HADS), parent-self-efficacy (PMP-SE)[243], satisfaction and empowerment (subscale on parent participation, EMPATHIC-N)[244], and impaired parent-infant bonding (PBQ)[245] (see S4. Appendix for details on the characteristics of the questionnaires).

## Statistical analyses

### Sample size calculation

We performed a sample size calculation for the AMICA study for the primary outcome of neurodevelopment in preterm infants at 2 years of age corrected for prematurity (See S5. Appendix for details on the sample size calculation[236]). We included sufficient parents for our psychometric analyses, as we had 10 participant responses per item.[246]

### Dealing with non-applicable responses and missing data.

We used the proposed guidance as explained by Sterne *et al.* [158] for missing data and applied the multivariate imputation by chained equations (mice) procedure with parcel summary scores to missing data at the item level.[247] Imputed datasets were used for further analyses[248], including confirmatory factor analysis (CFA) and construct validity.[249] We performed sensitivity analyses for data considered missing if participants did not fill out a question, or if items were scored as “this was non-applicable”. For all datasets we performed 10 imputations and 50 iterations to obtain imputed datasets (see S6. Appendix for variables included in the missing data model). Convergence was checked graphically with stripplots for Domain 1, 2, 3, 4 and 6, and convergence plots for Domain 5. Pooled estimates for further analyses were derived applying Rubin’s Rules.[160,250]

### Structural validity

#### *Confirmatory Factor Analysis*

Confirmatory factor analysis was done on imputed datasets using structural equation modelling. We used diagonally weighted least squares (DWLS). The DWLS approach uses the weighted least squares (WLS) estimator with polychoric correlations as input to create the asymptotic covariance matrix.[251] We calculated the following fit measures: comparative fit index (CFI), Tucker-Lewis index (TLI), Root Mean Square Error of Approximation (RMSEA) and the (Standardized) Root Mean Square Residual (SRMR).[252]

#### *Internal Consistency*

We calculated composite reliability (CR) for each domain to assess internal consistency, as the CR is calculated from factor loadings and acknowledges the possibility of heterogeneous item-construct relations and estimates true score variance from the factor loadings resulting in more precision for multilevel confirmatory factor analyses than the commonly used Cronbach's alpha.[253] Desirable values for CR are between 0.6 and 0.9.[254]

### Construct Validity

#### *Distinctiveness between domains*

We analyzed construct validity by using the Average Variance Extracted and Heterotrait-Monotrait criterion.[254] First, we determined the Average Variance Extracted (AVE) which informs how closely each domain is related based on the item characteristics within each domain, the AVE should be greater than 0.05 to be acceptable.[254] To examine the distinctiveness between domains we performed Heterotrait-Monotrait (HTMT), a new method that measures a ratio of correlation.[254] The HTMT method has emerged as a discriminant validity method that has been shown to achieve higher sensitivity and specificity (99% and 97%) than the commonly used cross-loadings and Fornell-Lacker methods.[254] We set our threshold for the HTMT analysis at 0.85.[254]

#### *Total scoring*

Total scores per domain were obtained by summing scores for hypothesis testing. For Domain 1, 2 and 6 we calculated 0 for 'The nurse does this', 1 for 'The nurse and I do this together' and 2 for 'I do this independently' (minimum scores 0 to 22, 8 and 14 respectively), indicating the positive inherent relationship between participation and collaboration. We performed sensitivity analyses on non-applicable items, either transforming them to 0 (no participation in this item) indicating that parents did not participate or did not experience an item or to missing before multiple imputation (and thus rendering a 0,1, or 2 value after multiple imputation). For domain 3 and 4 'yes' was scored as 1, and 'no' as 0 (minimum scores 0 to maximum 3). For the domain Time Spent with Infant (3 items) we performed sensitivity analyses including the items as scored originally (minutes or hours of relevant items) or as quartiles (minimum 0 maximum 12). Quartiles were calculated in imputed datasets. A total participation score was obtained by summing all domain scores. Minimum total scores were 0 and maximum 62.

***Hypotheses testing***

We calculated Pearson correlation coefficients (rho) and associations for hypothesis testing. We set up 5 hypotheses. A priori, we hypothesized (Hypothesis 1) that the total score would have a negative correlation with parent well-being outcomes such as depression and anxiety, of -0.3 to -0.5, meaning that if parents were depressed or anxious, they would demonstrate lower active parent participation. Contrarily, Hypothesis 2 was that the total score would have a positive correlation with self-efficacy and satisfaction and empowerment, of +0.3 to +0.5. We used univariate linear regression analysis to compare groups and test for associations. We stated that (Hypothesis 3) the CO-PARTNER-tool would be able to discriminate between high and low parent presence (Domain 5) and participation (total score) within the trial on the effect of FICare in SFR on parent and infant outcomes.[236] Also, we anticipated (Hypothesis 4) that mothers would be more present (Domain 5) than fathers, as fathers in the Netherlands had on average 2-5 days of paternity leave, and resume to work quickly after birth during conduct of the study.[255] The last hypothesis (Hypothesis 5) was that parents who were more present (Domain 5), would participate more in daily care (Domain 1).

### Statistical packages and software

We used R for statistical analyses (version 3.6.1)[163] for missing data analysis the 'mice'-package[256], for confirmatory factor analysis the 'lavaan'-package and 'semTools'-package.[257,258] For all tests, a *p*-value of less than 0.05 was considered statistically significant.

## RESULTS

During the conduct of the AMICA study, 1213 preterm infants were assessed for eligibility. In total, 309 families were included, with 358 infants, 296 mothers and 263 fathers (Fig 1). One hundred and seventy-four out of 296 included mothers and 132 out 263 included fathers (response rates 58.8% and 50.2% respectively) filled out the questionnaire on parent participation and collaboration at NICU discharge of their infant and were included in this psychometric study (see S7. Appendix on parent responses to the CO-PARTNER tool). There were 233 infants within 205 families. The median gestational age of their infants was $33^{+3}$ weeks, and parents filled out the CO-PARTNER tool at a median postmenstrual age of their infants of $37^{+1}$ weeks. Baseline characteristics of the sample are outlined in Table 2.

**Figure 1. Flowdiagram of study**

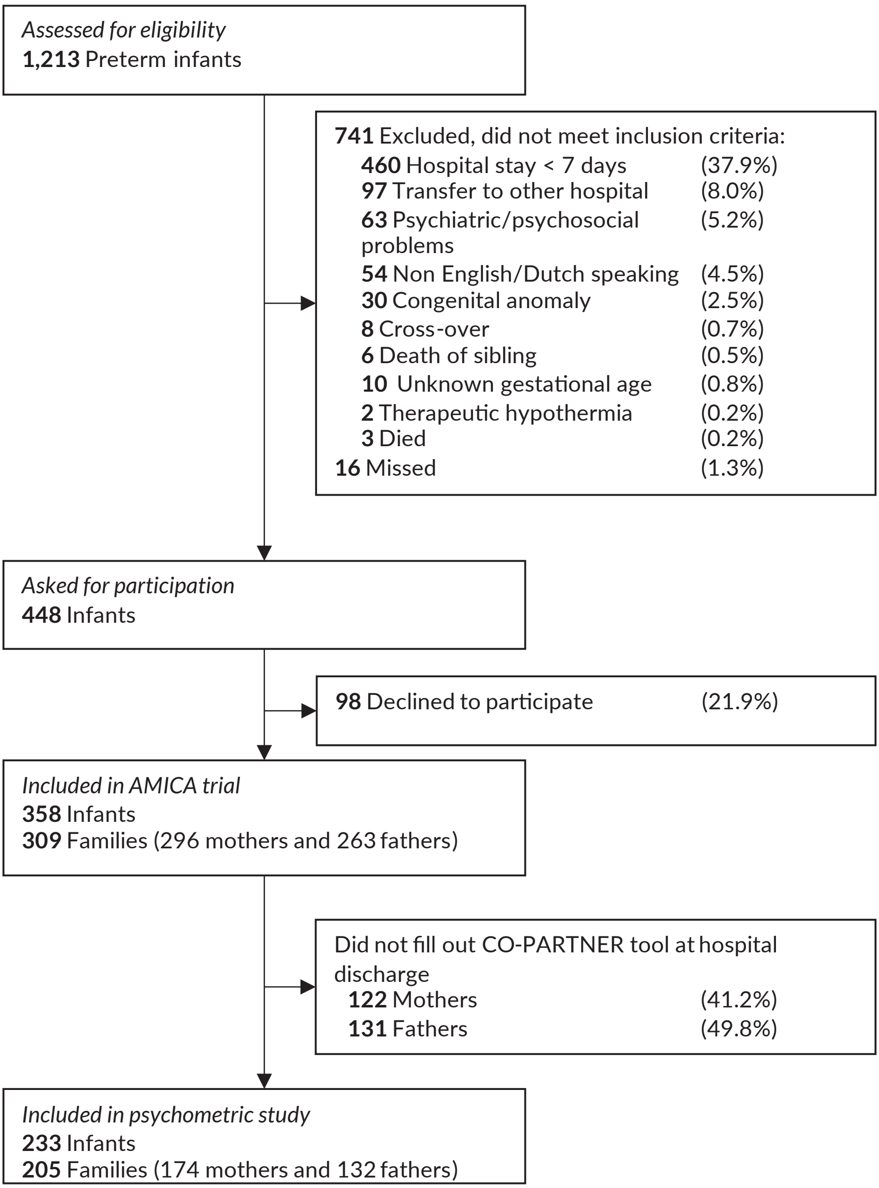


**Table 2. Baseline characteristics of the sample**

| | Included (n= 306 parents) | Missing (n (%)) |
|---|---|---|
| Mothers (n (%)) | 174 (56.9) | 0 |
| Admitted to FICare setting (n, (%)) | 157 (51.3) | 0 |
| Gestational age of infant at birth (weeks$^{+days}$, median (IQR), range (min- max)) | $33^{+3}$, ($31^{+0}$ - $34^{+6}$), ($24^{+5}$ - $36^{+6}$) | 0 |
| Postmenstrual age of infant at discharge to home (weeks$^{+days}$, median (IQR)) | $37^{+1}$ ($36^{+4}$ - $38^{+0}$) | 0 |
| Age (years, mean (SD)) | 34.4 (4.7) | 7 (2.3) |
| Higher education level (n, (%)) | 273 (89.2) | 14 (4.6) |
| Employed (n, (%)) | 259 (84.6) | 14 (4.6) |
| Work hours per week (mean (SD)) | 38 (7.4) | 2 (0.7) |
| Identifies with Dutch background (n, (%)) | 270 (88.2) | 9 (2.9) |
| Attended FICare sessions (n, (%)) | 64/157 (40.8) | 27 (8.8) |
| Supported by child psychologist during NICU stay (n, (%)) | 73 (23.9) | 42 (13.7) |
| Intends to raise child with partner (n, (%)) | 277 (90.5) | 15 (4.9) |
| Single parent (n, (%)) | 8 (2.6) | 15 (4.9) |
| First child upbringing (n, (%)) | 209 (68.3) | 13 (4.2) |
| Level of experienced stress during pregnancy (scale 1-5) (mean (SD)) | 2.2 (1.2) | 9 (2.9) |
| Level of experienced stress during birth (scale 1-5) (mean (SD)) | 2.8 (1.3) | 12 (3.9) |
| Anxiety and depression score at discharge (median, IQR) | 7 (4 - 12) | 23 (7.5) |
| Self-efficacy score at discharge (mean, SD) | 63 (8.9) | 29 (9.5) |
| Parent NICU stress score at discharge (total, mean (SD)) | 47.0 (23.6) | 23 (7.5) |
| Impaired parent-infant bonding score at discharge (median, IQR)) | 8 (4 - 13) | 13 (4.2) |
| Parent participation in EMPATHIC-N score (median, IQR) | 5.6 (5.1 - 6.0) | 10 (3.3) |

n: number, FICare: family integrated care, NICU: neonatal intensive care unit, SD: standard deviation

**Structural validity**

Three items were removed, and included items highly correlated with each other ( “Keep track of defecation of my child” and “Keep track of urination of my child”, transformed into “Keep track of output (urination and defecation) of my child”) and two items were deemed redundant in the analysis phase by the author group (“Walking a small round with my child if it is permitted” and “On average, how many minutes did you perform skin-to-skin per day?”). A total of 31 items were used in CFA. The fit parameters demonstrated good to moderate fit, CFI and TLI were 0.923 and 0.914, respectively, RMSEA 0.030 (90%CI: 0.021; 0.037), and SRMR (0.129). Factor loadings for domains are described in Table 3. Sensitivity analyses for missing data, revealed that model fit was better without transforming the non-applicable items to missing (see S8. Appendix for sensitivity analyses). The overall model fit increased if the domain *Time Spent with Infant (Domain 5)* was scored with quartiles.

The domains *Acquiring Information* (Domain 3) and *Parent Advocacy* (Domain 4) were initially included and evaluated as one domain (Advocacy). CFA revealed low factor loadings of *Acquiring Information* items to the overall domain of Advocacy. Post-hoc, better loadings were achieved when items were within the domain of *Acquiring Information*.



Factor loadings were 0.508 or higher in *Daily* Care (*Domain 1*, range 0.508-1.003). Within *Medical Care (Domain 2)* factor loadings ranged between 0.399 and 0.591. *Acquiring Information* (*Domain 3)* and *Parent Advocacy* (*Domain 4*) had overall good representation and items within the domain on *Time Spent with Infant (Domain 5)* loaded all above 0.7. The *Closeness and Comforting the Infant* domain showed overall factor loadings equal to or above 0.65, three items were low (between 0.487-0.566). The three lower items were, “Soothe my child during a painful procedure (for instance drawing blood)”; “Skin to skin contact”; and “Comfort my child whenever he/she needs it”. CR scores were strong in *Daily Care* (Domain 1, CR: 0.934), *Acquiring Information* (Domain 3, CR: 0.745), *Parent Advocacy* (Domain 4, CR: 0.855); *Time Spent with Infant* (Domain 5, CR:0.839) and *Closeness and Comforting the Infant* (Domain 6, CR: 0.871). CR within participation in *Medical Care* showed results just outside desirable ranges (Domain 2, CR: 0.558, see S9. Appendix for CR scores).

**Construct validity**

The Average Variance Extracted and HTMT demonstrated strong construct validity and distinctiveness of domains (see S10. Appendix for construct validity and distinctiveness outcomes). The direction of correlation between total and domain scores met our prespecified hypotheses (Fig 2 and S11. Appendix for outcomes of hypotheses testing).

**Table 3. Factor loadings after confirmatory factor analysis**

| Domain | Factor loading | Standard Error |
|---|---|---|
| Domain 1. Daily Care | | |
| 1. Bath my child/clean my child with a washcloth. | 0.508 | 0.058 |
| 2. Change my child's diaper. | 1.003 | 0.046 |
| 3. Feed my child (breast or bottle). | 0.681 | 0.068 |
| 4. Change my child's clothing. | 0.862 | 0.061 |
| 5. Get my child out of the incubator/cradle. | 0.640 | 0.084 |
| 6. Give my child medication. | 0.714 | 0.044 |
| 7. Weigh my child. | 0.652 | 0.043 |
| 8. Keeping track of output (urination and defecation) of my child | 0.775 | 0.033 |
| 9. Measure the temperature of my child. | 0.777 | 0.040 |
| 10. Keep track of my child's weight. | 0.775 | 0.033 |
| 11. Keep track of drinking and my child's feeds. | 0.790 | 0.031 |
| Domain 2. Medical Care | | |
| 12. Give tube feeding to my child. | 0.537 | 0.071 |
| 13. Look at my child's monitor and handling accordingly (e.g. stimulating during a bradycardia). | 0.424 | 0.079 |
| 14. Regulate the visiting of others to my child. | 0.591 | 0.093 |
| 15. Participate in the daily rounds with the doctor. | 0.399 | 0.072 |
| Domain 3. Acquiring Information | | |
| 16. Did you ask health care professionals information on the health of your child? | 0.84 | 0.198 |
| 17. Did you ask the health care professionals for information about your child for times when you were not present? | 0.584 | 0.167 |
| 18. Did you talk with another parent about your experiences? | 0.671 | 0.117 |
| Domain 4. Parent Advocacy | | |
| 19. I stood up for my child; I told somebody to do something in the care of my child. | 0.775 | 0.071 |
| 20. I stood up for my child; I told somebody NOT to do something in the care of my child; I gave boundaries | 0.747 | 0.064 |
| 21. I gave an explanation on the daily routines of my child to a health care professional. | 0.913 | 0.070 |
| Domain 5. Time Spent with Infant | | |
| 22. On average, how many hours per day were you present in the hospital with your child? | 0.946 | 0.122 |
| 23. On average, how many hours per day do you have contact with your child? | 0.98 | 0.128 |

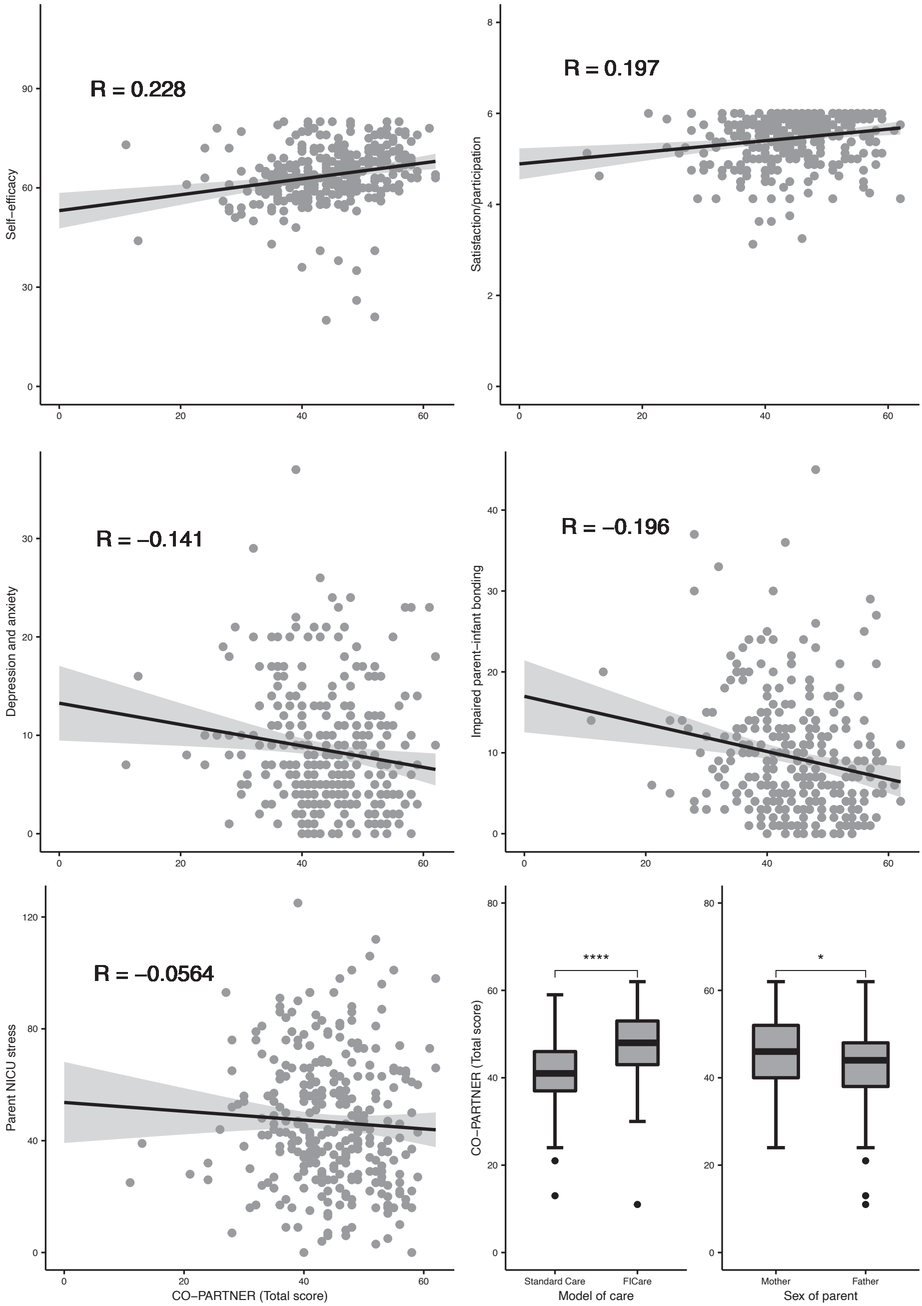
**Table 3. Factor loadings after confirmatory factor analysis (continued)**

| Domain | Factor loading | Standard Error |
|---|---|---|
| 24. On average, how many hours per day were you really close with your child? | 0.799 | 0.132 |
| Domain 6. Closeness and Comforting the Infant | | |
| 25. Hold/rock/cuddle my child. | 0.943 | 0.057 |
| 26. Comfort my child whenever he/she needs it. | 0.511 | 0.102 |
| 27. Kangaroo care / skin to skin contact. | 0.487 | 0.066 |
| 28. Be together with my child, be close with my child. (intimate time). | 0.566 | 0.095 |
| 29. Be together with my child (be present). | 0.995 | 0.048 |
| 30. Soothe my child during a painful procedure (for instance drawing blood). | 0.653 | 0.055 |
| 31. Recognize my child's signals. | 0.665 | 0.064 |

Negative correlations were present between total and domain scores on the CO-PARTNER tool with depression and impaired parent-infant bonding (Hypothesis 1). No correlations were found between the CO-PARTNER tool and parent NICU stress (total and domain scores). We found positive correlations for total and domain scores between parent participation and parent self-efficacy and parent satisfaction and empowerment (Hypothesis 2).

We confirmed our Hypothesis 3 that parents in the FICare group participated more, they had significantly higher total CO-PARTNER total scores (beta 6.020, 95%CI 4.144; 7.895, $p<0.0001$). Also, parents in FICare had higher subdomain scores than parents in the standard care group (including *time being present*, Domain 5), except for Domain 3 (*Acquiring Information*, see S11. Appendix). Likewise (Hypothesis 4), mothers had higher CO-PARTNER scores than fathers (beta 2.103, 95%CI 0.084; 4.121, $p=0.041$). Overall, parents who were present more (*Domain 5)* participated more in daily care (Hypothesis 5, *Domain 1*, beta 0.390, 95%CI +0.240; + 0.540, $p<0.0001$, see S11. Appendix for outcomes of hypothesis testing).

**Figure 2. Results of hypothesis testing**




Legend: Scatterplot and boxplot values are shown from the first imputed dataset. Correlation coefficients and significance are pooled outcomes from all imputed datasets. r: correlation coefficient (Pearson's rho)

## DISCUSSION

To our knowledge, this is the first study to perform rigorous instrument development and psychometric testing methodology to develop a measure of parent participation and inherent collaboration with healthcare staff in neonatal care. The six domains of this tool explicitly measure parents' participation and collaboration with care providers in their unique roles in care provision, leadership, and connection to their infant.

The psychometric evaluation demonstrated good content, construct and structural validity of the CO-PARTNER tool to the construct of parent participation in neonatal care. Overall, it was able to measure our pre-specified hypotheses. However, the factor loadings within Domain 2 (*Medical Care)* were not as desirable as we had hypothesized beforehand. This domain represents areas of care that are associated with hospital unit specific tasks and might contain items that parents were not familiar with (yet), insufficiently coached into, or in which nurses were not comfortable supporting parents in. There might also be individual preferences or variations to what extent parents want to participate in medical care. Parent participation in medical care is rapidly evolving and a new area of neonatal care that needs to be further explored. Specifically, the item on daily rounds should be studied more carefully as parents' desired role could be different from their actual role, possibly explaining the low factor loading within that domain.[226] Nevertheless, from a FICare perspective, parent active participation in daily rounding is key and therefore should be incorporated in the tool. Equally, the closeness and comforting items were loading satisfactory but not excellent. These questions have been formed with a collaborative component (in the item generation phase) when in fact the collaboration between parents and health care professionals might not be a relevant component for these items.

The CO-PARTNER tool encompasses elements of parent participation such as time spent with the infant, closeness with infant, collaboration and competencies in daily care activities that have been previously measured separately.[116,134,186,187,231] The CO-PARTNER tool included fathers from initial conception, which provided insight into their specific needs and support to feel comfortable and competent in caring for their baby. In contrast to previous tools, our newly developed tool incorporates collaborative features explicitly describing and incorporating the process of collaboration between parents and health care professionals within daily and medical care and decision-making for hospitalized neonates, in alignment with the construct to be measured.[70] Above, one of the main strengths is, that the tool was developed in close collaboration with parents, ensuring face and content validity. The tool was also acceptable and

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feasible for parents to fill out, with an average missingness in items of 2.3%, with 4 items >5% missingness and a maximum of 8.9%.

The findings from this study should be considered in light of its limitations. First, the CO-PARTNER tool is unable to distinguish between different kinds of collaboration, as that would increase the data collection burden. However, collaboration details can be explored within the context of trusting relationships between nurses and families. Together they could view results of the CO-PARTNER tool and consider the parent development towards performing activities independently as an examination of their collaborative processes during NICU hospitalization of their infant. Second, learning is not explicitly assessed with the tool. For instance (learning how to) feed a preterm or sick neonate can be technically challenging and parents develop skills over time.[259] The answer option “the nurse and I do this together” can be seen as a proxy for a learning scale, eventually resulting in parents feeling competent to do this independently. As the level of learning is different from the level of collaboration the tool is unable to measure learning processes directly. Another limitation is that the directions of correlations between the total participation score met our pre-specified hypotheses but were not strong. This could be due to the fact that the constructs for which we assessed the correlation were different. The correlation between depression and participation is expected to be much weaker than the correlation of the scores of the CO-PARTNER tool with another patient participation instrument, but this was not assessed within this study as no such tool was available. Within this psychometric study, we did not adjust for clustering within families but included fathers and mothers as separate individuals. Therefore, the possibility of non-independence of a couple’s responses cannot be ruled out[260] and should be explored in future studies. Also, parents who completed the tool were highly educated, and therefore future studies should include a more diverse sample of mixed levels of educated parents to validate our results.

The CO-PARTNER tool can be used to support quality improvement by health organizations, practitioners, and care specialists working within various NICU settings and with different models of (parent-partnered) care. This tool could potentially be used for benchmarking across and comparing settings. All items included in the CO-PARTNER tool can be performed by parents and this should be fully supported by units, as is advocated by parent representatives and the WHO.[78,261] With CO-PARTNER scores parents can provide actionable quantitative data on the level of parent participation in care, with lower scores suggesting more tasks performed solely by health care professionals without participation of parents. Equally, the CO-PARTNER tool can potentially enable comparison of parent-partnered care practices and to study (health) outcomes in infants and their parents through, for instance, mediation analysis.[262]

For clinical practice we envision that there is no summing of total scores, as the measure is intended to be a guide in understanding each parent's unique style of caring and participation and identify gaps in the culture of the unit. One could consider adding open-ended free-text questions to allow participants to explain some difficulties in their own words. However, for research and benchmarking between units total scoring can be meaningful; measuring parent participation in total or within subdomains can inform if interventions are needed to ameliorate family care practices. By measuring parent participation, researchers and parents can identify which collaborative practices are occurring in the NICU, which items are deemed not applicable by the parents, and subsequently work together to develop individualized strategies for improving parent participation rather than simply reporting quantity and types of tasks completed by parents.

Future research should focus on use of the tool in different settings (for instance in level 3 units), different countries, different intercultural contexts (for instance immigration, language or lower levels of education) and different resource settings (for instance in units relying on care delivery by families out of necessity), and with parents of infants with a wider range of diagnoses to determine if further adaptation is needed to account for context. It would also be interesting to evaluate the inter-rater-reliability between perspectives of parents and nurses on the items in this tool, which could enable an assessment of nurses' ability to collaborate with parents and enable parents' participation and tailor education programs further if deemed insufficient for parents or health care professionals. Likewise, the CO-PARTNER tool could be studied to evaluate progress within parents (beginning and end of hospital stay) or to evaluate changes in parent participation and collaboration after implementation of education programs for parents and health care professionals. Above, analyses of non-applicable items and their meanings related to unit culture could be studied further, preferably in mixed-method research understanding qualitative features of hospital care culture.

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## CONCLUSIONS

The CO-PARTNER tool is able to assess parent participation and the collaborative process between parents and health care professionals in the NICU for research and in care. The CO-PARTNER tool, developed on the basis of participation theory and with parent engagement design methods, can reignite health organizations' motivation toward researching, monitoring and implementing parent-delivered and parent support interventions in the NICU. The tool could serve as a standard measurement for parent-partnered interventions in the neonatal care unit.

Chapter 7

# **The functions of adequate communication in the neonatal care unit:** A systematic review and meta-synthesis of qualitative research

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# ABSTRACT

**Objective**
To assess the main functions of parent-provider communication in the neonatal (intensive) care unit (NICU) and determine what adequate communication entails according to both parents and health professionals.

**Methods**
A systematic review and meta-synthesis of qualitative research. PubMed, Ebsco/PsycINFO, Wiley/Cochrane Library, Ebsco/CINAHL, Clarivate Analytics/Web of Science Core Collection, and Elsevier/Scopus were searched in October-November 2019 for records on interpersonal communication between parents and providers in neonatal care. Title/abstract screening and full-text analysis were conducted by multiple, independent coders. Data from included articles were analyzed using deductive and inductive thematic analysis.

**Results**
43 records were included. Thematic analysis of data resulted in the development of the NICU Communication Framework, including four functions of communication (1. building/maintaining relationships, 2. exchanging information, 3. (sharing) decision-making, 4. enabling parent self-management) and five factors that contribute to adequate communication across these functions (topic, aims, location, route, design) and, thereby, to tailored parent-provider communication.

**Conclusion**
The NICU Communication Framework fits with the goals of Family Integrated Care to encourage parent participation in infants' care. This framework forms a first step towards the conceptualization of (adequate) communication in NICU settings.

**Practice implications**
Findings can be used to improve NICU communication in practice, in particular through the mnemonic TAILORED.

# 1. INTRODUCTION

Each year, approximately 15 million infants are born preterm (before 37 weeks' gestation). This amounts to ten percent of all infants worldwide[263] Preterm infants are born in the late second or third trimester of pregnancy, when organ systems are not fully developed yet. Preterm infants often need prolonged support, e.g. for breathing, nutrition, and regulation of body temperature. They are prone to complications like infections, intracranial hemorrhages, visual and hearing problems, and severe bowel problems (necrotizing enterocolitis) and their mortality rates are high. In the long-term, preterm infants often need continued medical care, e.g. for lung, cardiac, or neurologic problems, and their risk of delayed psychomotor development is increased. The lower the gestational age, the more support infants need and the higher their risk for long-term adverse outcomes.[264–266] Specialized medical care for preterm infants can be provided in the neonatal (intensive) care unit (NICU). Neonatal care is organized into four different levels (levels 1-4), corresponding to the complexity of care offered (see Table 1).[267]

**Table 1. Levels of neonatal care**

| Level of care | Description |
|---|---|
| Level 1<br>*Well-born nursery* | Postnatal care to stable term newborn infants as well as to infants born 35–37 weeks' gestation who remain physiologically stable. Stabilize infants' who are ill or born at < 35 weeks' gestation until transfer to a higher level of care |
| Level 2<br>*Special care nursery* | Medium to high complex neonatal care<br>Level I capabilities plus postnatal care for infants born ≥ 32 weeks' gestation and weighing ≥ 1500 grams who have physiologic immaturity or who are moderately ill with problems that are expected to resolve rapidly and are not anticipated to need subspecialty services on an urgent basis. Step-down unit from Level III. Stabilize infants born < 32 weeks' gestation or weighing < 1500 grams until transfer to a higher level of care. Brief mechanical ventilation possible. |
| Level 3<br>*NICU* | Level II capabilities plus provision of sustained life support and comprehensive care for infants born critically ill, before < 32 weeks' gestation, or weighing < 1500 grams. Access to pediatric medical subspecialists, advanced imaging techniques, and different forms of respiratory support. |
| Level 4<br>*Regional NICU* | Highly specialized neonatal intensive care<br>Level III capabilities plus capabilities to provide surgical repair of complex congenital or acquired conditions and access to full range of pediatric medical (surgical) subspecialists and pediatric anesthesiologists onsite. |

Based on the American Academy of Pediatrics[267]

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The NICU is a distressful environment for parents of preterm infants. Along with the concerns parents have for their infant's health and survival, they often experience the NICU as an unfamiliar, dauntingly complex, and frightening environment.[268] Throughout their infants' admission to the NICU, parents interact with various health care professionals, including neonatal physicians, neonatal nurses, social workers, physical therapists, speech therapists, and providers from other medical disciplines. These professionals not only provide care to preterm infants, but can also help parents to get acquainted with the NICU environment, to better understand their infants' medical status, and to make the transition to becoming independent caregivers at home. Throughout this process communication is pivotal. Several studies show that adequate communication between health care professionals and parents in the NICU contributes to parents' satisfaction with care and diminishes their stress levels.[269–271] More so, research shows that good communication ensures that parents feel more involved in the care of their child and, reversely, that poor communication can lead them to withdraw from the NICU and its staff, thereby hampering parent-infant attachment.[272–274] As such, it is important to understand what it entails for providers to *adequately* support parents during admission of their preterm infant to the NICU by means of interpersonal communication.

Over the past decades, the Family Integrated Care (FICare) model has received increasing attention in neonatal care.[89,215,275] FICare starts from the assumption that, ideally, parents and providers should work together in an equal partnership to foster parent-infant closeness, increase parents' participation in care, and ultimately improve short and long-term outcomes for preterm infants and their parents.[55,56,90] FICare consists of four core pillars: (1) the *NICU environment*, promoting a shift from open-bay wards to single family rooms to enhance parents' feeling of safety and privacy and increase parent-nurse collaboration;[276,277] (2) *psychosocial support*, improving parents' coping and allowing them to engage with their infant;[90,269] (3) *staff education and support*, encouraging training of NICU staff on how to help parents become more involved in practical care activities (e.g., feeding, diaper changes, skin-to-skin care);[55,89,278] and (4) *parent education*, offering training to parents to allow them to independently care for their infant upon discharge.[275,279] Across the four pillars of FICare, parent-provider interaction plays a significant role. Yet, to date, a systematic overview of the precise role and functions of communication within family integrated care is lacking. Moreover, it is unclear what 'good' parent-provider communication precisely entails.

In this review, we therefore systematically explore, synthesize, and analyze the literature on parent-provider communication in the NICU (level 2-4). Thereby, we focus specifically on medium to intensive neonatal care, excluding studies conducted in the well-born nursery (level 1). In this study we aim to: (1) assess the *main functions* of parent-provider communication in the NICU and (2) determine what *adequate* interpersonal communication in NICU settings encompasses, according to parents as well as providers. Defining interpersonal health communication as the (direct, non-mediated) verbal and non-verbal interaction between providers and patients, we use the Framework for Patient-Centered Communication by Epstein and Street as our starting point.[280,281] This framework has been developed to explore the relationships between different aspects of interpersonal communication and outcomes in oncology settings, yet fits well with the family-integrated approach to communication in neonatal care. As such, we seek to contribute to theoretical conceptualizations of NICU communication and – ultimately – to improve the quality of parent-provider interaction in practice.

# 2. METHODS

This systematic review and meta-synthesis of qualitative research is part of a larger endeavor to systematically search and analyze the literature on the *functions* of (adequate) parent-provider communication and its *effects* on parent-related outcomes during NICU admission. The present study includes only qualitative studies, as it seeks to synthesize parents' and providers' told perspectives ('narratives') on the functions of NICU communication. The search strategies for the overall project as well as both quantitative and qualitative findings pertaining to communication *effects* are reported elsewhere.[282] This review is reported in accordance with the PRISMA statement.[283] The review protocol is registered with PROSPERO (CRD42020150218).



## 2.1 Data collection

The literature search for the overall project was conducted in October-November 2019 by a medical information specialist (JK) and included indexed terms and free-text words for 'neonatal intensive care unit', 'parents', 'participation', and 'communication' or 'decision making'. The following databases were searched: PubMed, Ebsco/PsycINFO (23 October), Wiley/Cochrane Library (24 October), Ebsco/CINAHL, Clarivate Analytics/Web of Science Core Collection, and Elsevier/Scopus (28 November). No restrictions on language or publication date were imposed.

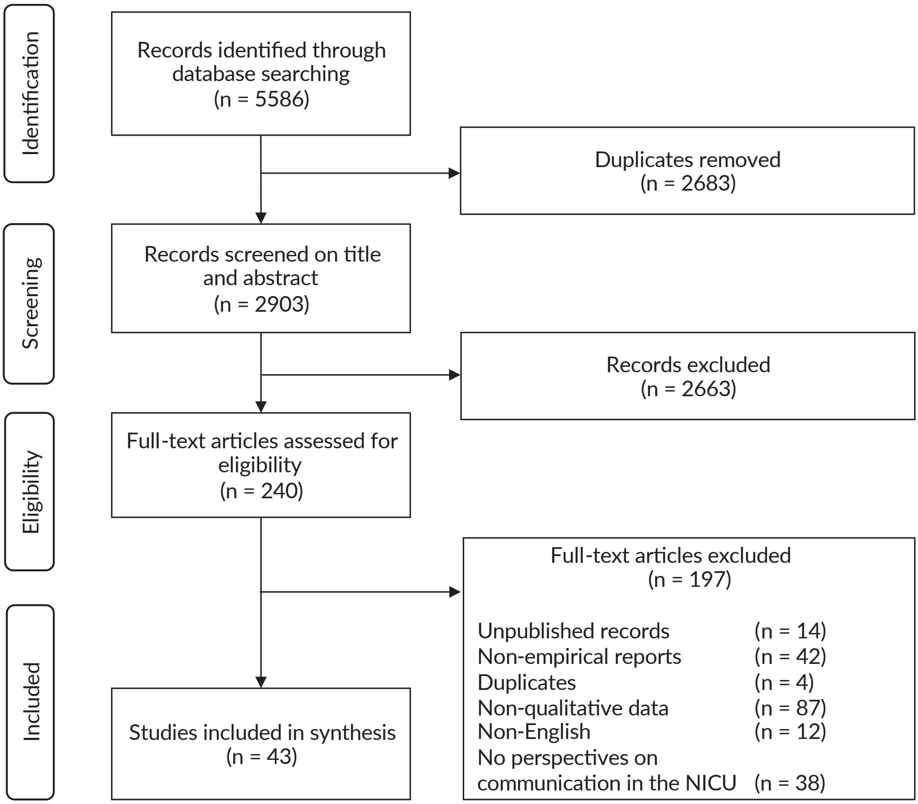
The initial search yielded 5586 records, from which 2683 duplicates were removed. The remaining 2903 records were uploaded in Rayyan QCRI.[284] Title/abstract screening was conducted by two independent coders (NV, NL), representing the medical and parent-communication perspective. All conflicts were resolved through discussion involving a third coder (AvK). Because inter-rater reliability for abstract/title analysis was fair to moderate (Cohen's kappa: 0.40) – likely due to the 'fuzziness' of the concept 'communication' – in case of doubt, records were included for full-text analysis. This resulted in 240 records for full-text assessment. Full-texts were retrieved via the library services of VU Amsterdam and OLVG Amsterdam.

Full-texts were assessed by WW and NL, applying the following inclusion/exclusion criteria: records had to be published in English and report on original, empirical, qualitative research focused on relevant stakeholders' perspectives on parent-provider communication in the NICU. Unpublished (e.g. abstracts, theses, posters), non-empirical (e.g. research protocols, reviews, editorials, opinion pieces), quantitative, and non-English records were excluded. Also, records reporting on interprofessional communication or parent-provider communication prior to or following admission to the NICU were excluded. Records concerning the development of communication resources such as decision-aids, websites, or parent education, as well as studies on cultural or linguistic barriers between parents and providers and the importance of interpreters were excluded, due to our focus on direct, rather than mediated, forms of interpersonal communication. Inclusion and exclusion criteria can be found in Table 2. For an overview of the search see Figure 1.

**Table 2. Inclusion and exclusion criteria for title/abstract screening and full-text analysis**

| | Inclusion criteria | Exclusion criteria |
|---|---|---|
| Type of participants | Parents of admitted infants to the NICU (level 2–4), neonatal physicians, neonatal nurses, and other stakeholders involved in parent-provider communication. | Parents and neonatal health professionals in a level 1 NICU, and before or after a level 2–4 NICU admittance. |
| Phenomena of interest | Interpersonal neonatal parent-provider communication | Interprofessional health professional communication; parent-provider communication prior to or following admission to the NICU; development of communication resources; and cultural or linguistic barriers between parents and provides. |
| Context | NICU admission | Outside NICU admission |
| Type of studies | Published records, original empirical research, qualitative records | Unpublished records (e.g. abstracts, theses, posters), non-empirical studies (e.g. research protocols, reviews, editorials, opinion pieces), and quantitative records. |
| Language | English records | Non-English records |

**Figure 1. Flow diagram of systematic review.**




## 2.2 Data extraction and analysis

To systematically extract and organize data from full-text records, a data extraction sheet was used (available upon request). The sheet included meta-data (e.g., authors, publication year) and methodological aspects (e.g., study setting, sample, NICU level, analytic methods). Data on communication functions were extracted and analyzed by WW and NL applying the procedures described by Finfgeld-Connet.[285] Data was extracted from the results sections only. Findings reported in discussion sections were excluded, to avoid extracting interpretations rather than data. Direct quotes from interviews or focus groups were not extracted to avoid bias. Data were analyzed using combined deductive and inductive thematic analysis.[286] Relevant findings from included records were deductively categorized in the extraction sheet, according to the communication functions described by Street et al.: fostering relationships (building trust and report between providers-patients), information exchange (allowing providers and patients to share knowledge and insights), responding to emotions (helping patients cope with difficult circumstances), managing uncertainty (helping patients interpret uncertain medical scenarios), decision-making (making appropriate decisions), and enabling self-management (helping patients independently manage health-related problems).[280,281]. Inductive analysis allowed for new functions of NICU communication to emerge.

Subsequently, the data within each communication function was coded inductively by WW, focusing on aspects defining what constitutes 'adequate communication' according to relevant stakeholders. NL independently coded a subset of data (60%). Codes were discussed and a codebook was developed. Finally, remaining codes were categorized by WW and NL into themes within and across communication functions. Themes were discussed within the research team.

### 2.3 Quality assessment

The quality of each individual article was evaluated by two independent coders for, within the scope of the broader project, using the 16-item Quality Assessment Tool for Studies with Diverse Designs.[287] Detailed results are reported elsewhere.[282] In this study, no records were excluded based on the quality assessment.

### 2.4 Ethical considerations

The present study is part of IMPACT, a comprehensive research program on NICU Communication. This program was approved by the Science and Ethics Committee of the Vrije Universiteit Amsterdam (VCWE-2019-132). The Medical Ethical Committee of the Amsterdam UMC, location VUmc judged that IMPACT is not subject to the Medical Research Involving Human Subjects Act, thereby waiving the requirement for medical ethical approval (2019.596).

## 3. RESULTS

### 3.1 Study overview

The search yielded 43 studies reporting on functions of parent-provider communication in NICUs (N=61) worldwide. Findings represent the perspectives of N=965 parents, N=54 family members, and N=409 care professionals. Notably, three times as many mothers (N=689) were included compared to fathers (N=226). The same applied to the number of nurses (N=279) versus doctors (neonatologists, pediatricians, fellows, residents) (N=112). Included studies used unstructured or semi-structured interviews, focus groups, and open-ended questionnaires to collect data and all applied thematic analysis. Table 3 summarizes the overall study population. Table 4 provides an overview of all included studies.

**Table 3. Study population**

| | N |
|---|---|
| Parents | 965 |
| Mothers | 689 |
| Fathers | 226 |
| Not specified | 50 |
| Family members | 53 |
| Health professionals | 409 |
| Nurses | 297 |
| Neonatologists | 112 |
| Staff educators | 11 |
| Speech therapists | 1 |

### 3.2 NICU Communication Framework

Seeking to determine the main *functions* of parent-provider communication in the NICU, based on our synthesis of data we developed a new model, constituting an adaptation of Street's framework.[280,281] The NICU Communication Framework encompasses four main functions of communication in the NICU: (1) building and maintaining relationships, (2) exchanging information, (3) (sharing) decision-making between parents and providers, and (4) enabling parent self-management.

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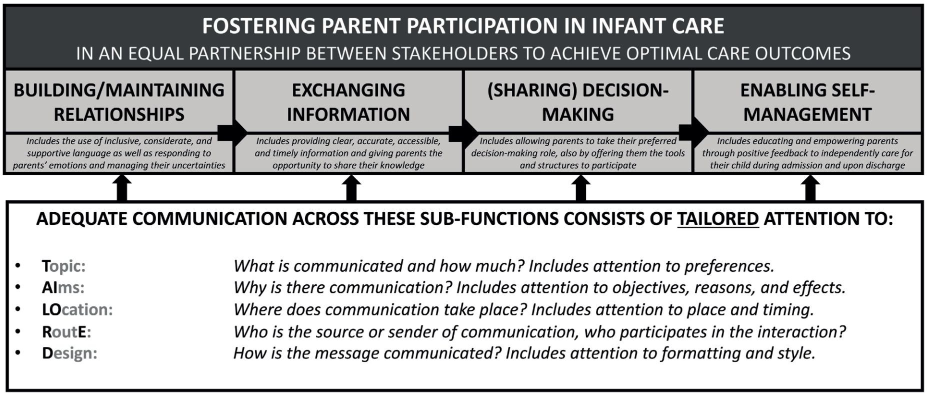
The NICU Communication Framework befits the unique context of neonatal care, in which providers communicate with parents rather than with patients, and medical care is often both acute and long-term. The four functions of the NICU Communication Framework contribute to Family Integrated Care in the NICU, as communicative interaction is considered to be ideally directed towards fostering parents' participation in infant care in an equal partnership with providers, to achieve the best possible outcomes of care – both during admission and following discharge. Notably, in the NICU Communication Framework the function *building and maintaining relationships* also encompasses providers responding to parents' emotions and managing uncertainty. As relationships between health care professionals and parents in the NICU appear to be built and maintained first and foremost through regulation of parents' emotions and distress and uncertainty management (e.g. about infants' prognosis), these functions from the original framework are clustered.

In the NICU Communication Framework, communication functions are ordered sequentially. Building supportive relationships between parents and providers is considered fundamental in order to effectively exchange information about infants' medical situation. This, in turn, allows parents to engage in decision-making about treatment and care plans in their preferred role, which finally is seen as a prerequisite to empower parents to take part in practical care activities and feel prepared to independently care for their child upon discharge. The NICU Communication Framework, thus, offers an ideal model which outlines the ordered functions communication may have in parent-provider interaction. Needless to say, practice may deviate from the theoretical ideal and in reality, functions may be achieved continuously, simultaneously, and sometimes in a different order.

Inductive analyses revealed five reoccurring factors that are important for *adequate* communication in NICU settings, across all functions of communication. These factors include providers' deliberate attention in their communication with parents to the (1) topic, (2) aims, (3) location, (4) route, and (5) design of the interaction. Prior to initiating interaction with parents, to ensure communication adequacy, providers should carefully consider the precise *topic*, or content, of what is going to be discussed (e.g. infant status, daily 'chit-chat', treatment information). Providers should determine the main *aim* of the conversation (e.g. informing, reassuring, or preparing parents). Furthermore, providers should think of the right *location* and timing of the communication (e.g., open ward or in a separate room, directly following admission or before discharge) and what is the proper *route* for communication. The route may include a choice between a (dedicated) nurse or a neonatologist, but also the option to offer written information or to conduct conversations via telephone. Finally, in the *design* providers should consider the communication style they want to use (e.g. objective and direct, empathic, coaching). Obviously, in considering these factors providers should take into account parents' preferences and allow room for their contributions, too. Together, the five factors of adequate communication form the acronym TAILORED, which can serve as a mnemonic for providers to memorize what it entails to adapt their communication to parents' situational and personal needs. The four communication functions and five factors of adequate communication, jointly constitute the baseline for the NICU Communication Framework (Figure 2). What it entails to adequately address each of the aforementioned factors across the four functions of communication, will be discussed next.

**Figure 2. NICU Communication Framework**




## 3.3 Functions and factors contributing to adequate NICU communication

### 3.3.1 Building and maintaining relationships

Building and maintaining positive relationships with parents includes responding to their emotions, fulfilling their supportive needs, and helping parents to manage uncertainties in their infant's care. Properly attending to this function of communication highly impacts parents' satisfaction with infants' care.[226,291] Analyses reveal that to build solid parent-provider relationships, parents want to discuss medical *topics* with their infants' neonatologists on a regular basis. This helps them to become more familiar with the NICU environment, to feel reassured, and more at ease to ask questions.[291,296,298,300,305,306,308,317,327] To develop good relationships with nursing staff, parents also particularly appreciate daily 'chit-chat' on topics that do not necessarily concern the infant's care:

*"Nurses who 'chatted' and conveyed a sense of partnership and equality were also frequently mentioned as supportive. Parents valued nurses who [...] were able to engage in conversations that recognized that there was 'life outside' the nursery."* [308]

In their endeavors to build relationships with parents, providers should *aim* to make parents feel comfortable in the NICU and feel more involved with their infant's care. They should strive to get to know the parents and respond to their personal needs.[290,294,296,299,300,305,308,314,323,327–329] Parents want to develop good relationships with providers to receive guidance and support during difficult times, but also to ascertain their infant receives the best of care possible.[294,299] Parents appreciate reassurance from providers, as this helps them mitigate their fears and overcome traumatic experiences.[296,302–304,306,308,321,324] Having regular conversations in a secluded *location* such as a single room increases parents' sense of privacy, their confidence to ask questions, and the idea that providers take the time to listen. This allows parents to show their emotions, in turn enabling providers to support parents during difficult conversations.[311] In terms of communication *route*, parents prefer to build and maintain trusting relationships with dedicated nurses and neonatologists, rather than encountering many different professionals.[292,294,298] Conversation *designs* that can be used by providers to build a good relationship include showing concern, understanding, and empathy to parents as well as demonstrating their professional experience.[291,300,305,308,309,311,316,324] Parents particularly appreciate communication that is respectful, compassionate, caring, and genuine to build and maintain good relationships with provider.[301,310,311]

*"Families reported that their relationships with staff were central to their satisfaction with care. They judged these relationships based on whether providers were physically available, compassionate, and genuine in their interactions. This was true across specialties, but parents most frequently acknowledged the care and dedication of nursing staff."* [291]

### 3.3.2 Exchanging information

Exchanging information involves a continuous sharing of knowledge between parents and providers throughout infants' admission. Parents and providers can exchange information on *topics* regarding the medical condition, treatment plans, and possible outcomes of the infant.[226,288,297,302,304,306,307,309,311,313,314,321,324] In order to allow for optimal information exchanges, providers should consistently *aim* to help parents understand why certain care is provided, what this entails, and which treatment options are available to enable parents to participate in their infants' care.[297,309–311] Providers should explicitly encourage question-asking as well as information sharing. This increases parents' feeling of involvement, helps them to engage in decision-making, and reduces their anxiety:

*"Participants reported that the anxiety of mothers was relieved when the nurses constantly informed them about their newborn's condition and treatment, such that the mothers understood the health status of their babies."* [321]

Also here, parents prefer regular information exchanges in secluded *locations* to ensure that bad news, or unexpected or complex information is conveyed privately.[226,288,291,297,302–304,306,307,309,311,313,314,321,324] To ensure information provision is consistent, a designated nurse and neonatologist should be assigned throughout admission (*route*). Receiving conflicting information or different opinions from different providers may leave parents confused.[289,290,295,298,307,310,312,314,317] During and following information exchanges with neonatologists, nursing staff can assume a supporting role by helping parents understand the information provided.[226,288,296,304,309,310,313,326] Conversation *designs* that can be used by providers to ensure adequate information exchanges include being direct, consistent, clear, and thorough.[288,291,296,299,301,310,314,315,319,320] Furthermore, in providing information providers must avoid jargon and carefully consider the proper amount of information given, to reduce parents' distress and dissatisfaction.[291,296,297,302,304,305,308,310,311,313,318,324]

*"The fathers perceived disagreement between staff members as upsetting and confusing. Conflicting information and conflicting opinions among the staff about, for example, limits for alarms from medical equipment, were perceived as very negative, and physicians' use of medical terminology impeded the information flow."* [314]

### 3.3.3 (Sharing) decision-making

Throughout hospitalization, parents and providers continuously have to make decisions about treatment. Depending on parents' preferences as well as medical circumstances over the course of admission, decisions are made by providers, parents, or jointly. Analyses show that, generally, parents prefer to be involved when medical expertise is not required, when the risks involved are high, or when they consider issues as 'normal parenting' decisions (*topics).*[171,315,320]

*"Preferences for greater parental control were associated with high perceived risk, high parental knowledge about or personal experience with the decision, involvement of foreign bodily fluids such as blood, and similarity to decisions that parents perceived to be part of the normal parental role."* [171]

When parents feel decisions require medical expertise, they prefer to delegate decision-making to providers. Nonetheless, they want to be informed about the decision-making process.[171,315,318,319,325] Thus, providers should *aim* to involve parents in their preferred decision-making role, as this gives parents a feeling of being taken seriously and provides them with a sense of control.[226] However, this aim is not always achieved:

*"[Parents] wanted inclusion in conversations about their child's care and engagement in decision making, but often struggled to find a way to be involved."* [292]

Parents emphasize that also in order to engage in decision-making, a secluded *location* to talk to providers is necessary.[304] Access to decision-aids, such as pamphlets explaining conditions, assists parents in the decision-making process *(route).*[310,311,319,324]. In terms of communication *design*, providers must seek to adapt their communication style to parents' personal needs concerning their involvement in decision-making.[171,291,295]

*"Being listened to in a genuine and consistent manner was also very important to many of the parents. They wanted to have a voice and be taken seriously when it came to identifying changes in their baby's condition and decisions about care including strong beliefs some parents held about how early care impacts on future outcomes of their babies."* [320]

#### 3.3.4 Enabling parent self-management

Enabling parent self-management concerning their infants' care is key during admission and after discharge. Providers can enable parent self-management by offering education on *topics* such as skin-to-skin care, breast-feeding, and changing diapers, or by teaching them about the use of medical equipment around the NICU.[290,292,299,306,308,314,320] Practical preparation for discharge is important, too.[290,306,313,314,322] More so, psychological care, through positive feedback, is deemed crucial:

*"Nurses also verbalized the importance of making parents feel positive about their ability to provide direct care to their infant. There was a genuine desire to assist parents to 'parent' and to make sure they were left feeling positive about doing the activity again or 'trying' again. Two nurses discussed possible differences between mothers and fathers stating fathers often 'required more encouragement' than mothers to be involved in caring for their baby."* [308]

Providers should *aim* to increase parents' confidence and empower them to participate in their infant's care during admission and to prepare them for discharge and their time home.[292,294,306,314,322] In terms of *location*, education may take place at infants' bedside or in group sessions. However, timing is particularly important. Providers should ensure timely education and discharge planning, to avoid overwhelming parents with information too close prior to discharge and to maintain parents' confidence in their parenting skills once at home.[290,302,313,314,322] In order to adequately self-manage infant care once providers become less available, parents need to have access to reliable information resources (*route*).[292,306,313] These may include parents' notes from conversations with providers, but also supplementary written materials which can be explained by providers during discharge preparation.[291,310,312–314] When encouraging parents to participate in care, providers should carefully observe parents' responses and explicitly encourage question-asking (*design*). This reduces the risk of misunderstanding and discouragement.[288,292,294,302,306,313,314]

*"When [parents] perceived that nurses were not fully engaged in 'helping' them with their parental role, they became disaffected and dissatisfied. Working in a 'harmonious' way with nurses was challenging for parents when they perceived nurses to be 'controlling'."* [308]

# 4. DISCUSSION AND CONCLUSION

## 4.1 Discussion

This systematic review and meta-synthesis provide a comprehensive overview of the role of interpersonal communication between parents and providers in NICU settings and offers practical insights into what it entails to communicate adequately within this context, according to both parents and health care professionals. This study has led to a first outline of the NICU Communication Framework, encompassing four *functions* of parent-provider communication: (1) Building and maintaining relationships, (2) exchanging information, (3) (sharing) decision-making, and (4) enabling parent self-management. The NICU Communication Framework provides a contextualization and refinement of Epstein and Street's Framework for Patient-Centered Communication in oncology settings.[280,281] Our data showed that several functions included Epstein and Street's Framework had to be merged and no new functions emerged in the ideal model. The new NICU Communication Framework fits within FICare practices, which seek to foster parents' participation in infant care in an equal partnership with providers, to achieve the best possible outcomes of care – both during admission and following discharge.[89,90,328] We therefore believe it is important that insights from the NICU Communication Framework are adopted and integrated in FICare, to further improve parent involvement in infant care.

Notably, there is a sequential order between the different functions of communication in the NICU Communication framework, with the fulfillment of each function being fundamental for optimally achieving the next. Thereby, enabling parents to self-manage in their infants' care, inherently builds on having good parent-provider relationships, adequate information exchange, and involving parents in decision-making in their preferred role. As such, neonatal providers can be seen to carry a double task of being responsible for infant care as well as for empowering mothers and fathers in their parental roles. This requires unique skills from NICU staff. Within FICare, several educational programs have been developed to facilitate providers in striving to improve (health) outcomes of infants and parents through greater parent-participation.[69,275,330] We recommend such programs to incorporate a communication component.

In addition to the functions of NICU communication, our analyses also show what constitutes *adequate*, tailored communication between parents and providers.

The importance of tailored communication and its potential cost-effectiveness in healthcare was already discussed two decades ago.[331] More recently, research showed that providing tailored communication results in better medical adherence, better health outcomes, and a higher quality of life after recovery.[280,281,332–336] However, what it exactly means to tailor communication to patients' needs, is by no means evident from the literature. The present study shows that, within NICU settings, across all four functions of interpersonal communication, adequate interaction entails that providers seek to consistently pay attention to the *topic*, *aims*, *location*, *route*, and *design* of their communication – thereby adapting their communication to parents' needs in a given situation. Providers may use the acronym TAILORED as a mnemonic to remember the five factors of adequate communication.

Interestingly, while the ways in which the five factors take shape differ for each of the communication functions, the results consistently show that across all functions parents need providers to ensure that interaction takes place in private settings – enclosed locations where parents have more privacy, feel at ease to ask questions, and where they can participate in discussions and in care. This result resonates with findings of van Veenendaal et al., who demonstrate that single family rooms contribute to parent participation during NICU admission.[147] This implies that for optimal communication to take place, NICUs should consider implementing single family rooms rather than open wards. Taken together, these insights are beneficial for understanding what is needed to facilitate tailored and family-integrated parent-provider communication in neonatal care.[337]

It may seem contradictory that, while the NICU Communication Framework promotes an equal partnership between parents and providers, it only formulates specific requirements (i.e. TAILORED-factors) for providers on how to communicate with parents. After all, a true partnership would entail that parents contribute equally to the interaction and, consequently, that requirements should be formulated for them, too. However, while parents' role in family-integrated care is equally important to the roles of healthcare providers, their role is also unique. Given the emotionally challenging circumstances for parents imposed by the NICU, we believe it is important that providers take the lead using the TAILORED-factors in order to conjure an equal partnership and individualized communication with parents throughout their presence in the NICU.

Notably, there is a great asymmetry with regard to the participants included within the data of this meta-synthesis. Fathers are heavily underrepresented in the data. However, studies have shown that fathers experience also high levels of stress, respond differently to situations in the NICU compared to mothers, and use different coping mechanisms throughout admission.[338,339] It is thus important to further explore fathers' perspectives to provide adequate support during this difficult period and after.[340] The same asymmetry exists between providers. Nurses' perspectives are overrepresented compared to neonatologists. The role of nurses as primary informants for parents is increasing.[341] The overrepresentation of nursing staff may also be a reflection of the size of the nursing team compared to the number of doctors in a neonatal ward. However, as the results show, certain communication functions are primarily fulfilled by physicians and parents require adequate interaction from both types of providers. As such, more attention should be paid to neonatologists' role in parent-provider interaction.

The results of this meta-synthesis have to be interpreted in light of some limitations. First, inclusion for full-text analysis was limited to English articles, resulting in the exclusion of 12 articles and inducing a bias towards Anglo-Saxon neonatal cultures. Although our analyses were comprehensive and thematic saturation was reached at all levels, we cannot be certain that excluded records would have revealed different insights concerning parents' and providers' preferences and needs for NICU communication in and between different cultures. Such information could help to overcoming communication barriers when cultural differences between parents and providers arise.[342,343] Second, we cannot ascertain whether participants and NICUs overlap between different studies, thereby potentially affecting our interpretations. Third, the use of Epstein and Street's Framework for Patient-Centered Communication may have biased our analyses towards finding similar communication functions for NICU interaction. Yet, in addition to deductive analyses, we also conducted inductive thematic analyses to allow for new functions to arise – which was not the case. A strengths of this study concerns the involvement of a multidisciplinary research team, including health communication researchers, neonatologists, experts of family-integrated care, parent representatives, and a medical information specialist. Independent analysis of all records and data by multiple coders enhances the validity of our findings. Finally, in our analyses, we purposefully included the views of parents as well as health care professionals, thereby warranting a full-blown picture of what adequate NICU communication entails.

### 4.2 Conclusion

While the past years have seen increasing attention to the communicative interaction between parents and providers in the NICU, to date – and to the best of our knowledge – no systematic review has been performed to aggregate all findings concerning the *functions* of parent-provider interaction and the characteristics of *adequate* NICU communication. This review sought to include the perspectives of both parents and providers and, through a meta-synthesis, develop a new theoretical framework for family-centered and tailored communication in neonatal care. The resulting NICU Communication Framework is uniquely applicable to the neonatal context and can support further refinement and implementation of the Family Integrated Care Model. The present results and framework can be used in health communication research that seeks to improve parent-provider interactions in the NICU. Upon empirical testing, the NICU Communication Framework can be used to develop effective interventions to enhance tailored communication between parents and providers during their presence in the NICU – ultimately resulting in greater parent participation and better health outcomes. Also, the *effects* of communication on parent-related outcomes should be carefully explored in order to build a more comprehensive framework.[282]

**Practice Implications**

The findings have direct practical relevance, as providers may use the framework and the TAILORED acronym – which serves as a mnemonic – to refine their own communication strategies when engaging in dialogues with parents. TAILORED includes all factors for providers that play an important role in fulfilling the different functions of parent-provider communication in a given situation. Upon further testing of the NICU Communication Framework, NICUs may adopt this acronym to positively impact their staff's communication strategies. For instance, the acronym and short explanations of the different factors may be printed on a pocket-sized card to serve as a tangible reminder prior to engaging in conversations with parents. Also, staff training sessions based on the NICU Communication Framework may be useful to improve practice. Whilst the framework is thus still under development, it has the potential to significantly affect both research and practice.

**Table 4. Study characteristics of included records**

| Authors, year | Purpose or aim | Geographic location | N NICU(s) | NICU level | Sample | Methods |
|---|---|---|---|---|---|---|
| Able-Boone, Dockecki, & Smith, 1989[288] | Investigated parents' and healthcare providers perspectives of their communicative interactions when a seriously ill infant is treated in an intensive care nursey. | USA | 1 | 2 - 3 | 16 mothers<br>11 fathers<br>15 nurses<br>15 neonatologists | Open-ended and focused interviewing using an ethnographic interview method, data coding and analysis |
| Arockiasamy, Holsti, & Albersheim, 2008[289] | Understanding the experiences of fathers of very ill neonates in the NICU. | Canada | 1 | 3 | 16 fathers | Semi-structured interview, thematic analysis |
| Axelin, Outinen, Lainema, Lehtonen, & Franck, 2018[226] | Explored the dynamics of neonatologist parent communication and decision- making during medical rounds in a level three neonatal intensive care unit. | Finland | 1 | 3 | 15 mothers<br>7 fathers<br>2 neonatologists | Ethnographic approach with semi-structured interviews, thematic analysis |
| Aydon, Hauck, Murdoch, Siu, & Sharp, 2018[290] | Explore the experiences of parents with babies born between 28–32 weeks' gestation during transition through the neonatal intensive care unit and discharge to home. | Australia | 1 | 3 | 20 mothers<br>20 fathers | Semi-structured interview, thematic analysis |
| Baughcum et al., 2017[291] | Examined the EOL experience of families in the NICU, and methodological issues, particularly the lack of standardized measures, have limited our understanding of how to optimize care. | USA | 1 | 4 | 42 mothers<br>28 fathers | Semi-structured interview, thematic analysis |
| Berman et al., 2019[292] | To explore the parent perspective on discharge home from the neonatal intensive care unit (NICU). | USA | N/A | 2 - 3 | 14 mothers<br>1 father | Ethnographic approach with semi-structured interviews, thematic analysis |

**Table 4. Study characteristics of included records (continued)**

| Authors, year | Purpose or aim | Geographic location | N NICU(s) | NICU level | Sample | Methods |
|---|---|---|---|---|---|---|
| Bracht, O'Leary, Lee, & O'Brien, 2013[86] | To develop, implement, and evaluate a parent education and support program that enhances family-integrated care in a Canadian neonatal intensive care unit (NICU). | Canada | 1 | 3 | 39 mothers<br>11 staff educators | Evaluating structured interview |
| Brinchmann, Førde, & Nortvedt, 2002[293] | To generate knowledge about parents' participation in life-and- death decisions concerning their very premature and/or critically ill infants in hospital neonatal units. The question is: what are parents' attitudes towards their involvement in such decision making? | Norway | N/A | N/A | 19 mothers<br>16 fathers | Semi-structured interview, thematic analysis |
| Broom et al., 2017[294] | To first describe parents' and staff perceptions of the benefits of each component of the FICare program and second to explore parents' and staff perceptions of the FICare program in an Australian NICU. | Australia | 1 | 3 | 4 mothers<br>1 grandparent<br>8 nurses | Focus groups, thematic analysis |
| Bruns & McCollum, 1999[295] | What roles do mothers assume in communication with NICU medical professionals participating in their infants' care? | USA | 1 | 3 | 7 mothers<br>1 nurse | Semi-structured interview, thematic analysis |
| Bruns & McCollum, 2002[296] | Examine the perspectives of mothers, nurses, and neonatologists on the importance and implementation of NICU practices related to caregiving, information exchange, and relationships within the context of family-centered care. | USA | 6 | 3 | 55 mothers<br>122 nurses<br>18 neonatologists | questionnaires with open-ended questions, thematic analysis |

**Table 4. Study characteristics of included records (continued)**

| Authors, year | Purpose or aim | Geographic location | N NICU(s) | NICU level | Sample | Methods |
|---|---|---|---|---|---|---|
| Cox & Bialoskurski, 2001)[297] | (1) Identification of factors associated with the provision of information that may facilitate and hinder family attachment. (2) Exploration of problems associated with communication caused by family and, in particular, mother–infant separation, while the infant is being cared for in a NICU. | UK | 1 | 3 | 32 mothers<br>10 family members | Unstructured interviews and focus groups, thematic analysis |
| Falck, Moorthy, & Hussey-Gardner, 2016[298] | Examine provision of Palliative Care as experienced by mothers and healthcare providers (HCPs) of NICU patients with life-threatening illnesses. | USA | 1 | 4 | 6 mothers<br>5 nurses<br>1 neonatologist | Semi-structured interview, thematic analysis |
| Fenwick, Barclay, & Schmied, 2000[299] | To improve care provided to parents of infants in het neonatal nursey by understanding and explaining the experience of mothering in level II nurseries. | Australia | 2 | 2 - 3 | 31 families | Unstructured interviews, thematic analysis |
| Fenwick, Barclay, & Schmied, 2001[300] | Explores the use of `chat' or `social talk' as an important clinical tool that can assist nurses achieve family-centred care in neonatal nurseries. | Australia | 2 | 2 - 3 | 28 mothers<br>20 nurses | Semi-structured interview, thematic analysis |
| Flynn & McCollum, 1993[301] | To obtain parents' opinions about the types and sources of formal support available for their family during their child's hospitalization, as well as the perceived adequacy of formal support and gaps in services. | USA | 1 | 3 | 6 mothers | Open-ended focused interview, thematic analysis |

**Table 4. Study characteristics of included records (continued)**

| Authors, year | Purpose or aim | Geographic location | N NICU(s) | NICU level | Sample | Methods |
|---|---|---|---|---|---|---|
| Geetanjli, Manju, Paul, Manju, & Srinivas, 2012[302] | To determine and assess the loss and grief response, and perceived needs of parents who are having their newborns in neonatal care units. | India | 2 | 3 | 16 parents | Semi-structured interview, thematic analysis |
| Guillaume et al., 2013[303] | to explore parents' perception of these first interactions and to identify the actions of caregivers that help or hinder its development. | France | 3 | 3 | 30 mothers<br>30 fathers | Semi-structured interview, thematic analysis |
| Harvey, Nongena, Gonzalez-Cinca, Edwards, & Redshaw, 2013[304] | To explore parental information and communication needs during their baby's care in the neonatal unit with a focus on brain imaging and neurological prognosis. | UK | 1 | 3 | 13 mothers<br>5 fathers | Semi-structured interview, thematic analysis |
| Hendriks & Abraham, 2017[305] | To explore parental attitudes and values in the end-of-life decision-making process of extremely preterm infants (gestational age < 28 weeks). | Switzerland | 1 | 3 | 12 mothers<br>8 fathers | Semi-structured interview, thematic analysis |
| Hinton, Locock, Long, & Knight, 2018[306] | To understand the experiences of parents of infants who required surgery early in life. To identify messages and training needs for the extended clinical teams caring for these families—including pediatric surgeons, neonatologists, nurses, obstetricians, midwives and sonographers. | UK | 1 | 3-4 | 33 mothers<br>11 fathers | Semi-structured interview, thematic analysis |

**Table 4. Study characteristics of included records (continued)**

| Authors, year | Purpose or aim | Geographic location | N NICU(s) | NICU level | Sample | Methods |
|---|---|---|---|---|---|---|
| Ichijima, Kirk, & Hornblow, 2011[307] | Examines sources of parental stress in the two neonatal intensive care units (NICUs) located in New Zealand and Japan and explores how cultural norms of NICU care environments influence parental stress-related experiences and nursing support. | New Zealand / Japan | 2 | 2 - 3 | 30 mothers<br>17 fathers | Quantitative questionnaire, thematic analysis |
| Jones, Taylor, Watson, Fenwick, & Dordic, 2015[308] | To describe parents' and nurses' perceptions of communicating with each other in the context of the special care nursery. | Australia | 2 | 2 - 3 | 27 mothers<br>4 fathers<br>12 nurses | Semi-structured interview, thematic analysis |
| Kavanaugh, Moro, & Savage, 2010[309] | To describe nurse behaviors that assisted parents to make life-support decisions for an extremely premature infant before and after the infant's birth. | USA | 3 | 3 | 40 mothers<br>14 fathers<br>29 nurses<br>42 neonatologists | Semi-structured interview, thematic analysis |
| Kavanaugh, Savage, Kilpatrick, Kimura, & Hershberger, 2005[310] | To describe decision making and the decision support needs of parents, physicians, and nurses regarding life support decisions made over time prenatally and postnatally for extremely premature infants. | US | 2 | 3 | 6 mothers<br>2 fathers<br>2 nurses<br>6 neonatologists | Semi-structured interview, thematic analysis |
| Kodjebacheva et al., 2017[311] | Investigated strategies for effective health communication in the NICU. | USA | 1 | 3 | 6 mothers<br>2 fathers<br>17 nurses<br>3 neonatologists | semi-structured interview and focus groups, thematic analysis |
| Lemmen, Fristedt, & Lundqvist, 2013[312] | To describe parents' experience of information and communication mediated by staff nurses before and during KC at neonatal wards. | Sweden | 3 | 3 - 4 | 12 families | Semi-structured interview, modified content analysis on the basis of Graneheim and Lundman |

**Table 4. Study characteristics of included records (continued)**

| Authors, year | Purpose or aim | Geographic location | N NICU(s) | NICU level | Sample | Methods |
|---|---|---|---|---|---|---|
| Lemmon, Donohue, Parkinson, Northington, & Boss, 2016[313] | To characterize the parental experience of communicating with clinicians about TH and neonatal encephalopathy. | USA | 1 | 4 | 20 parents | Semi-structured interview, thematic analysis |
| Modé, Mard, Nyqvist, & Blomqvist, 2014[314] | To explore fathers' perception of information received during their infants' care at a neonatal intensive care unit (NICU). | Sweden | 2 | 3 | 8 fathers | Semi-structured interview, thematic analysis |
| Lundqvist, Nilstun, & Dykes, 2002[315] | To examine and illuminate mothers' experiences and perceptions of the care given to them at neonatal clinics while facing the threat and the reality of losing their baby. | Sweden | 1 | 2-3 | 16 mothers | Semi-structured interview, hermeneutic phenomenological method |
| Lupton & Fenwick, 2001[316] | To investigate the ways in which women with hospitalized newborn infants construct and practice motherhood. | Australia | 2 | 2-3 | 31 mothers<br>20 nurses | Semi-structured interview, thematic analysis |
| Lyndon, Wisner, Holschuh, Fagan, & Franck, 2017[317] | To describe parents' perspectives and likelihood of speaking up about safety concerns in the NICU and identify barriers and facilitators to parents speaking up. | USA | 1 | 3 | 14 parents | Semi-structured interviews, thematic analysis |
| Mburu, Wardle, Joolay, & Densmore, 2018[318] | To discuss how technology design processes with and for mothers of preterm infants who are susceptible to stress look like in practice. | Namibia | 1 | 3 | 15 mothers<br>10 nurses<br>5 neonatologists | Semi-structured interviews, thematic analysis |
| Payot, Gendron, Lefebvre, & Doucet, 2007[319] | To explore empirically how parents and neonatologists engage in the decision to resuscitate a baby at the threshold of viability. | Canada | 1 | 3-4 | 8 mothers<br>4 neonatologists | Semi-structured interviews, thematic analysis |

**Table 4. Study characteristics of included records (continued)**

| Authors, year | Purpose or aim | Geographic location | N NICU(s) | NICU level | Sample | Methods |
|---|---|---|---|---|---|---|
| Petty, Jarvis, & Thomas, 2019[320] | Focuses on what students and staff can learn from parents about what they feel is important to make their experience better. | UK | N/A | 3 | 19 mothers<br>4 fathers | Narrative interviews, thematic analysis |
| Phuma-Ngaiyaye & Welcome Kalembo, 2016[321] | To investigate the strategies for supporting maternal–newborn bonding for mothers whose neonates were admitted to an intensive care unit at a tertiary hospital in Malawi. | Malawi | 1 | 3 | 10 mothers<br>5 nurses | Semi-structured interviews, thematic analysis |
| Raffray, Semenic, Osorio Galeano, & Ochoa Marín, 2014[322] | To explore Colombian health care provider perceptions of barriers and facilitators to preparing families with premature infants for discharge home from the neonatal intensive care unit (NICU). | Colombia | 1 | 2 - 3 | 11 nurses<br>3 neonatologists<br>1 speech therapist | Semi-structured interviews, thematic analysis |
| Rodrigues, Uema, Rissi, Felipin, & Higarashi, 2019[323] | To understand the nursing team's perception regarding family centered care and its practice in the neonatal intensive care unit. | Brazil | 1 | 3 | 19 nurses | Semi-structured interview, thematic analysis |
| Russell et al., 2014[324] | Explores parents' views and experiences of the care for their very premature baby on NICU. | UK | 3 | 3 | 32 mothers<br>7 fathers | Semi-structured interview, thematic analysis |
| Silva & Osswald, 2010[325] | Sheds light on the views of health care providers and parents, and its results can inform and improve the decision-making process in these settings. | Portugal | N/A | N/A | 14 mothers<br>1 nurses<br>13 neonatologists | Semi-structured interview, thematic analysis |
| Uhl, Fisher, Docherty, & Brandon, 2013[326] | To describe parents' care experiences during hospitalization of their children to identify strategies that could improve the provision of patient and family-centered care (PFCC). | USA | 1 | 3-4 | 7 mothers<br>2 fathers | Semi-structured focus group & survey, thematic analysis |

**Table 4. Study characteristics of included records (continued)**

| Authors, year | Purpose or aim | Geographic location | N NICU(s) | NICU level | Sample | Methods |
|---|---|---|---|---|---|---|
| Weis, Zoffmann, & Egerod, 2015[327] | To explore how parents of premature infants experience guided family-centered care (GFCC), and (b) to compare how parents receiving GFCC versus standard care (SC) describe nurse-parent communication in the neonatal intensive care unit. | Denmark | 1 | 3 | 12 mothers<br>10 fathers | Semi-structured interview, thematic analysis |
| Weiss, Barg, Cook, Black, & Joffe, 2016[171] | To explore how characteristics of medical decisions influence parents' preferences for control over decisions for their seriously ill infants. | USA | 2 | 3 | 25 mothers<br>5 fathers | Semi-structured interview, thematic analysis |

# Chapter 8

# **Effects of parent-provider communication during infant hospitalization in the NICU on parents:** A systematic review with meta-synthesis and narrative synthesis

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# ABSTRACT

## Objective

To synthesize and analyse the literature on the effects of parent-provider communication during infant hospitalization in the neonatal (intensive) care unit (NICU) on parent-related outcomes.

## Methods

Systematic review with meta-synthesis and narrative synthesis. Databases (PubMed, PsycINFO, Cochrane Library, CINAHL, Web of Science, Scopus) were searched in October/November 2019. Studies reporting, observing, or measuring parent-related effects of parent-provider communication in the NICU were included. Study quality was assessed using the Quality Assessment Tool for Studies with Diverse Designs. Qualitative studies were meta-synthesized using deductive and inductive thematic analysis. Quantitative studies were analysed using narrative synthesis.

## Results

5586 records were identified; 77 were included, reporting on N=6960 parents, N=693 providers, and N=300 NICUs. Analyses revealed five main (positive and negative) effects of parent-provider interaction on parents' (1) *coping*, (2) *knowledge*, (3) *participation*, (4) *parenting*, and (5) *satisfaction*. Communication interventions appeared impactful, particularly in reducing parental stress and anxiety. Findings confirm and refine the NICU Communication Framework.

## Conclusions

Parent-provider communication is a crucial determinant for parental well-being and satisfaction with care, during and following infant hospitalization in the NICU.

## Practice implications

Providers should particularly consider the impact on parents of their day-to-day interaction – the most occurring form of communication of all.

# 1. INTRODUCTION

In the neonatal (intensive) care unit (NICU), parents continually engage in communication with healthcare staff. Communication between parents and providers serves important clinical goals. Through interaction information is relayed, consent is obtained, and decisions are made. In addition, communication can foster collaboration between parents and staff. Family-centred care models strongly encourage effective parent-provider communication, to provide care that fits individual families' needs and preferences.[4,83,84,344,345]

Studies exploring parents' NICU experiences confirm the importance of adequate parent-staff communication.[299,304,313,346–350] Parents value language that makes them feel emotionally supported, treated with empathy, and taken seriously.[278,294,299,348,349,351] They prefer clear, accurate, accessible, and timely information exchanges that allow them to share their knowledge.[289,314,348,349,352] Furthermore, studies show the importance of allowing parents to engage in decision-making in their preferred role, for example by using decision-aids or implementing family-centred rounds.[154,353–357] Finally, both parents and providers emphasize the need to empower parents to independently care for their infants during admission and upon discharge.[86,290,292,308,322] Taken together, these findings have previously led us to develop the NICU Communication Framework.[348,349] This framework outlines the four main functions of adequate parent-provider communication in neonatal care: (1) building and maintaining relationships, (2) exchanging information, (3) (sharing) decision-making, and (4) enabling parent self-management.

However, a systematic overview of the *effects* of NICU communication on parents is still lacking. This concerns effects that are self-reported (e.g., in interview studies and surveys) and observed (e.g., observational designs), as well as effects that have been established (e.g., in randomized trials). Across various healthcare domains, there is ample evidence that communication can affect, e.g., patients' understanding and memory of medical information[358–363], their adherence to treatment plans[364,365], their perceptions of their providers[359,360,365], and patients' physical and psychological wellbeing.[361,362,366] In neonatal care, it can be assumed that such positive communication effects are present, too. At the same time, communication may also cause adverse outcomes, such as misunderstandings, conflict, or stress. The present study seeks to methodically map the effects (reported, observed, and established) of parent-provider communication during infant hospitalization in the NICU (level 2-4, Appendix A[367]) on parent-related outcomes through a systematic literature review of qualitative and quantitative data.

# 2. METHODS

## 2.1 Reporting standards

This systematic review is reported in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA) statement[368], the Enhancing transparency in reporting the synthesis of qualitative research (ENTREQ)-checklist[369] for qualitive studies, and the Guidance for Reporting Involvement of Patients and the Public (GRIPP-2) short form.[211] The review protocol is registered with PROSPERO (CRD42020150218). Deviations from the protocol and all definitions used are reported in Appendices B and C.

## 2.2 Search strategy

Systematic searches were performed by an information specialist (JK) in the databases of PubMed and Ebsco/PsycINFO (up to 23 October 2019), Wiley/Cochrane Library (up to 24 October 2019), Ebsco/CINAHL, Clarivate Analytics/Web of Science Core Collection and Elsevier/Scopus (up to 28 November 2019). Searches included indexed terms and free-text words related to 'neonatal intensive care unit', 'parents', 'participation', and 'communication' (Appendix D). There were no restrictions on language, date, study type, or publication status.

## 2.3 Study selection, inclusion and exclusion criteria

Two researchers (NL and NvV) independently screened abstracts and, subsequently, assessed full-text articles for inclusion in Rayyan QCRI[370]. Coding conflicts were resolved through discussion, involving a third analyst (AvK). In case of doubt, full-text analysis was applied. Records in languages other than English were assessed in collaboration with native speakers. To determine interrater reliability, we calculated Cohen's kappa and specific agreement.[104]

Records were considered eligible for inclusion if they reported on scientific, empirical research –qualitative, quantitative, or mixed methods – studying the effects of communication between parents and healthcare providers during infants' NICU stay (level 2-4) on parent-related outcomes (Appendix E). We were interested in all self-reported, observed, and established effects of parent-provider communication. Records reporting data and outcomes of parent-infant or parent-professional dyads were also included: we then focused solely on parent-related outcomes. Prespecified effects included, but were not limited to, parents' cognitive (e.g., recall, understanding) and affect-oriented outcomes (e.g., trust, perceived physician credibility and caring), and effects on parents' psychological (e.g., stress, anxiety, attachment) and physical wellbeing (e.g., blood pressure), regardless of their timing (during hospital stay or follow-up). Also, adverse effects (e.g., misunderstandings, conflict, stress) were considered.

### 2.4 Data extraction

Data were extracted using a data extraction sheet (Appendix F). Data included meta-data (e.g., authorship, publication year); methodological aspects (e.g., design, setting, sample size, communication intervention/exposure, instruments, analytic approach); and reported communication effects (e.g., qualitative interpretations, statistical evidence). To avoid bias, data were extracted from results sections only and direct quotes from interviews of focus groups were not included.[285]

### 2.5 Quality assessment

We used the 16-item Quality Assessment Tool for Studies with Diverse Designs (QATSDD) to assess study quality.[287] This tool has good reliability and validity across study domains and is suited for the assessment of all study designs.[371] Quality of each record was independently assessed by four coders (NL, NvV, AvK, SvdS) after random allocation. Another coder (RL) provided training and acted as a judge in case of coding discrepancies. In case scores between the coders differed by less than one point, an average score was used as final rating. In case of larger or consistent discrepancies, RL checked the full-text article and decided on the final score.

### 2.6 Data analysis and statistical methods

A meta-synthesis of qualitative data was performed in Atlas.ti, using combined deductive and inductive thematic analysis.[208] Data were coded separately for reported positive and negative communication effects. First, references in the data to *communication* as well as the *effects* of communication were inductively coded by one coder (NL). The unit of analysis was a single utterance consisting of an independent clause or subclausal unit. Subsequently, the *communication codes* were deductively sorted into the four functions of interpersonal communication outlined in the NICU Communication Framework[349]: (1) building and maintaining relationships, (2) information exchange, (3) (sharing) decision-making, (4) enabling parent self-management. The *effect codes* were inductively grouped thematically into main themes and, when applicable, clustered into subthemes. A second coder (AvK) independently checked a subset of 20% of all data to ensure validity and rigor. All coding procedures were discussed among the interdisciplinary research team.



To analyse our quantitative data, we used the ‘meta’ and ‘metafor’ packages in R (version 3.6.1).[179] We assessed clinical, methodologic, and statistical heterogeneity in communication interventions to determine suitability for meta-analysis. As interventions appeared heterogenous and only few randomised trials were included, meta-analysis was not deemed appropriate and narrative synthesis was performed on all quantitative studies. We used guidance from the Centre for Reviews and Dissemination to provide full-text narrative review of our findings.[372]

### 2.7 Patient involvement

NL assumed a dual role of investigator-parent representative, being a mother to a daughter born at 26 weeks' gestation. Furthermore, three parent representatives provided feedback on our analyses and manuscript. Among these are the founders of a Dutch parent-support platform (Kleine Kanjers) and a parent-support organization for parents of preterm infants (Veerkrachtige Ouders).

### 2.8 Ethics approval

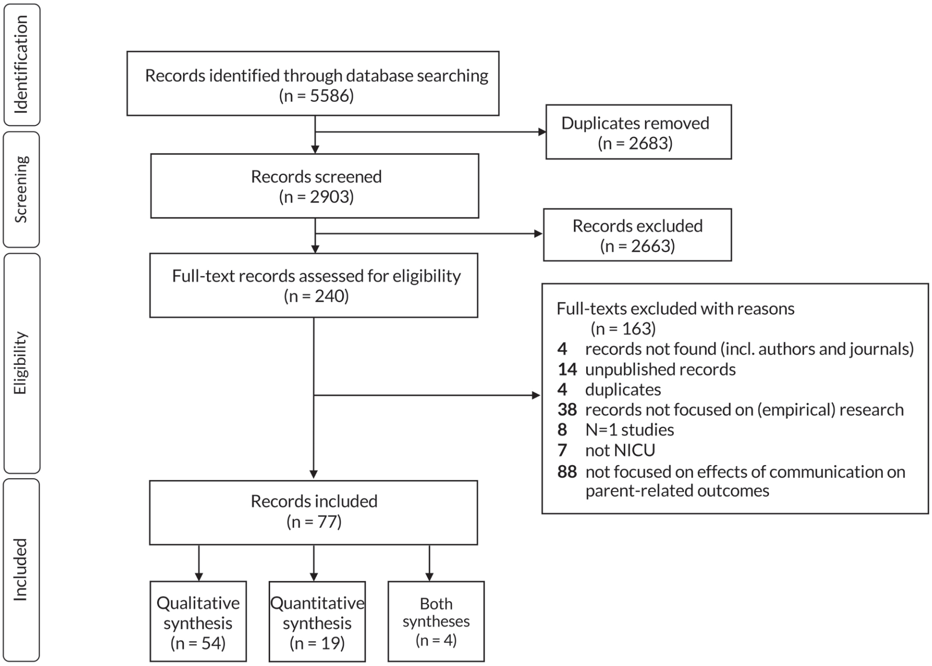
This study complies with the ethical guidelines of the Vrije Universiteit Amsterdam (VCWE-2019-132). The project was submitted for consideration to the Medical Ethical Committee of the Amsterdam UMC, location VUmc. The committee judged that the study is not subject to the Medical Research Involving Human Subjects Act (2019.596).

## 3. RESULTS

### 3.1 Search results

The search yielded 5586 records. 2683 duplicates were removed. Through title/abstract screening, 2583 records were excluded; 91 records were included by both independent coders and 229 conflicts arose. Inter-rater reliability for abstract/title analysis was fair to moderate (Cohen's kappa: 0.40; overall agreement 92.1%; positive specific agreement: 44.3%; negative specific agreement: 95.8%). Coders agreed about exclusion, but less so about inclusion due to the 'fuzziness' of the concept 'communication'. Following discussion, all conflicts were resolved. This resulted in 149 additional included records, arriving at 240 records for full-text analysis. During full-text analysis, 156 records were excluded by both coders, 70 records were included, and 14 conflicts arose. Inter-rater reliability for full-text screening was high (Cohen's kappa: 0.87; overall 94.1%; positive specific agreement: 91.1%; negative specific agreement: 95.6%). Following discussion, full agreement was reached and 7 more records were included, resulting in a total of 77 included records (Figure 1 and Table 1).

**Figure 1. Flowchart.**




### 3.2 Description of included records

Studies reported on data of N=6960 parents (N=3499 mothers, N=715 fathers) and N=693 professionals in approximately 300 NICUs around the world. Studies from Anglo-Saxon countries were dominant. Studies were published between 1989-2019, with about half published in the last five years. The majority of studies (N=64, 83.1%) reported on the communication between parents and providers in level 3 units. In total, N=47 qualitative records were included, N=19 quantitative records, and N=11 records reporting on mixed methods studies. With mixed designs, only those results were included that were relevant to our research question. This resulted in N=54 records included in the meta-synthesis, N=19 records for quantitative analyses, and N=4 in both. For qualitative studies, interviews were the most commonly used method (N=42), with thematic (content) analysis as the main analytic approach. Among the quantitative studies, there were 4 randomized controlled trials.

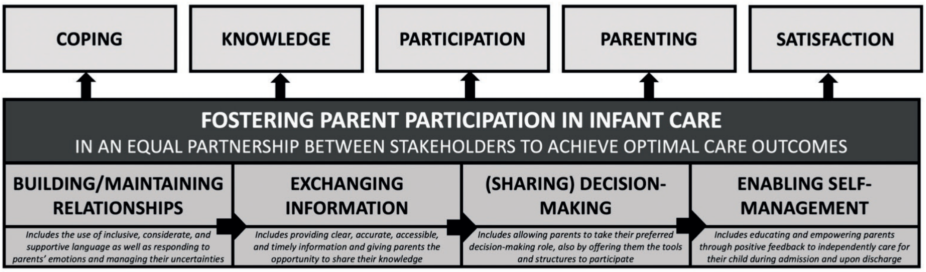
### 3.3 Quality assessment

Quality of included studies varied, with scores varying between 9/42 and 35/42 (Table 2). Study design did not seem to systematically influence quality rating. Limitations of included records related to missing evidence concerning sample size, inadequate justification for the analytic method, and a lack of patient/user involvement. In contrast, most studies provided a comprehensive literature review, clear description of objectives, and detailed account of the setting. No studies were excluded based on quality assessment.

### 3.4 Meta-synthesis and narrative synthesis

Meta-synthesis of qualitative data revealed five main outcomes of parent-provider interaction, including positive and negative effects on parents' (1) *coping*, (2) *knowledge*, (3) *participation*, in *communication* and in *care*, (4) *parenting*, in terms of *empowerment* and parent-infant *attachment*, and (5) *satisfaction* with overall care and relationships with providers, during and following NICU admission. These effects occur across all four functions of communication.[349] An overview of communication functions and their main effects is provided in Figure 2.

**Figure 2. NICU Communication Framework**




Quantitative studies reported on interventions across the four functions of communication (Table 3 and Appendix F). Within the function of *building and maintaining relationships*, three studies reported on communication interventions or exposures concerning supportive or inclusive communication.[269,397,408] Within the domain of *exchanging information*, four studies focused on ensuring information accessibility and allowing parents to share.[379,400,403,404] Seven studies described interventions or exposures on the function of *(sharing) decision making*, including formal structures to facilitate decision-making and decision aids.[154,186,356,373,378,385,386] *Enabling self-management* interventions included one study on discharge planning.[405] Finally, eight studies included an overall communication intervention. These focused on the implementation of Family Integrated Care (FICare)[55,56] or Guided Family-Centred Care[327], overall end-of-life communication[374], and parent-provider interaction in a broad sense.[347,384,387,390,401]

Below, the qualitative and quantitative evidence concerning the main effects of parent-provider interaction in the NICU is jointly reported, systematically linking parent effects to communication functions.

**3.4.1 Coping.** *"Both parents and nurses recognized that effective communication could reduce parents' stress"*[308]
Coping effects of communication included parents' ability to handle stress and deal with the infant's medical circumstances. Positive effects mentioned included parents feeling more at ease and comfortable within the NICU environment, improved emotional state, and stress reduction.[294,300,380,393] For instance, open and jargon-free information exchanges as well as parents' participation in medical rounds, during which parents can ask questions and decisions are made, increased parents' confidence and reduced their anxiety.[226,288,293,307,313,314,321,376] Also, supportive and empathic communication and positive nurse-parent relationships were frequently mentioned to alleviate parents' stress[226,298,300,309,380]:

*"Transparent communication that provided information in a personalized and sensitive manner facilitated development of trusting relationships and minimized maternal anxiety."*[298]

*"Nurses provided emotional support by (a) taking the time to listen, (b) forming a bond with the mother, (c) being non-judgmental of the parent lifestyle or decision choices, (d) offering hope, and (e) giving spiritual support. These behaviours helped mothers get through the emotionally charged nature of their experience and were done to decrease stress and to help them comprehend all of the information they needed in order to make decisions."*[309]

Also, enabling parents' self-management through effective discharge or transfer planning, seemed to have positively impacted parents' coping[298,308,381]:

*"Often the transfer happened quite quickly and there was not enough time for parents to be properly informed and have all their questions answered. The earlier and the more often that transfers were discussed the less distressing it was when it happened."* [381]

Negative communication effects reported by parents included feelings of anger, self-blame, guilt, and hopelessness.[291,352,380,381,399] They reported communication issues to cause anxiety and depression as well as difficulties adapting to, and accepting, the medical circumstances surrounding their infants' health.[299,322,352,377,380] Specifically, discordant or conflicting medical opinions among staff induced feelings of frustration[288,290,308,394]:

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*"Parents shared experiences where staff offered inconsistent or conflicting information which left them frustrated and confused."*[290]

Also, conflict and undermining and insensitive language – including "banter and nicknaming infants"[300,380,406] – were mentioned to cause extreme distress and anger, even making parents avoid the unit:

*"When this [being instructed] became 'being ordered' through the use of harsh tones and abrupt manners the situation was made all the worse. [...] In some situations the distress and anger felt by women were so great that they were unable to conceal their emotions. Covering up how they felt in an attempt to remain silent meant they must withdraw from the nursery."*[380]

Quantitative studies showed that coping significantly improved through interventions focused on, e.g., conveying empathy, the use of decision-aids, adequate discharge preparation, the introduction of care planning meetings, family centred rounds, and family-integrated care.[55,56,269,374,378,385,387,390,397,405] Overall, studies across all functions of communication reported lower stress and anxiety scores due to the described communication interventions.

**3.4.2 Knowledge.** *"Parents found simpler information was easier to understand and pass along*.**"**[313]
The effects of communication on parents' knowledge pertained to the extent to which parents understood their infant's situation and developed the skills to provide care. Positive communication experiences were mentioned to positively affect parents' recall of medical information and increased comprehension both in the short- and long-term.[291,292,314] Clear, timely, and reliable information improved parents' understanding and memory of their infant's medical status:

*"To learn and retain knowledge on how to care safely for their child while in the NICU, parents needed access to reliable sources of information, available when they were ready."* [292]

Notably, maintaining good relationships with providers changed parents' perceptions of the quality of the information received[309,314]:

*"If the first information given soon after the infant's birth was plain and concrete, the fathers found it easier to comprehend, otherwise they hardly remembered anything* [...] *Good personal chemistry between the staff and the father had positive effects on the perceived quality of the information."*[314]

Providers could positively influence parents' recall and understanding through opportunities and tools to prepare for, and engage in, medical rounds[294,311,389,407]:

*"Having reflected in advance [with a reflection sheet] helped the parents remember and bring up important issues that might not otherwise have surfaced."*[407]

Negative communication experiences, however, resulted in difficulties understanding, remembering, and processing information.[313,396] Parents reported feeling upset and uninformed when communication was unfriendly or inhibited mothering.[300,380] Also, language issues, information complexity and overload, untimely information, and medical jargon were mentioned as problematic[313,314,375,383,388,391,396,398,399,401]:

*"Nearly all parents remember feeling bombarded with technical information that confused the 'big picture'."*[313]

Quantitative studies showed that parents' knowledge increased significantly with the introduction of interventions concerning building and maintaining relationships, exchanging information, and shared decision making. Because of these interventions (such as care planning meetings, video conferencing, a baby diary, interdisciplinary family conferences, family centred rounds, decision-aids) parents felt more informed and their understanding of their baby's care increased.[154,356,373,374,379,385,403]

**3.4.3 Participation.** *"The mother's participation was supported by reciprocal communication."* [226]

Communication also impacted parents' participation, both in terms of communication and in practical care activities. Positive communication effects mentioned included parents experiencing the ability to ask questions, to express oneself, and to provide feedback.[294,389,391,407] Parents felt part of a team, as well as taken seriously.[294,381] Programs and tools focused on informing parents, inviting parents to share their thoughts, encouraging them to take part in medical decision-making, and counselling parents to become independent in managing their infants' health, seemed to positively affect parent participation in communication and care[226,288,294,314,346,391,395,402]:

*"The knowledge parents gained through individual bedside education and group information sessions [in FICare] was empowering, giving parents the confidence to actively participate in their infant's care and build a more trusting relationship with staff."*[294]

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*"Gathering information about their child's medical status and progress was a key coping strategy for nearly all parents. Many felt that it allowed them to be more effective participants in care and decision making."*[402]

Negative communication experiences, contrarily, were associated with a lack of partnership between parents and providers, feelings of being excluded from care and decision-making, a sense of being not valued or needed.[299,312,347,399] Only one study stated that parents do not want to be involved in decision-making.[325] Unpleasant interactions (unintentionally) caused parents to feel "stupid and incompetent"[308] and afraid to participate in discussions and confront health care professionals in case of conflict:

*"Parents found it difficult to muster the strength to confront nurses who had been insensitive to them or their infant."*[407]

In quantitative studies reporting interventions across all domains of communication, parents noted increased participation, except for the intervention focused on enhancing self-management, which did not measure participation as an outcome. When parents were involved in care planning meetings aimed at building relationships with staff, they also reported more involvement in shared decision-making.[397] When parents received information on the (medical) condition of their baby they felt more involved in their baby's care[404] and also family-centred rounds ensured that parents felt they could communicate effectively with staff and ask questions.[373] Overall communication strategies correlated positively with the feeling of being included in care[374], but when communication was not felt to be supportive this made parents feel less confident.[401]

**3.4.4 Parenting.** *"Communication with caregivers appeared decisive for fathers and mothers in feeling a bond with their child."*[303]

In terms of parenting, the communicative interaction between parents and providers affected mothers and fathers becoming empowered in their role as parents. Additionally, communication impacted parent-infant attachment, both positively and negatively. Reported positive effects of communication included, e.g., parents feeling confident and competent in taking care of their infant, assuming responsibility for their child, and considering themselves important experts.[226,294,310,355] Positive communication experiences were said to foster closeness and connectedness, and meaningful interactions between parents and infant.[297,303,314,321] Nursing staff who demonstrated empathy, sensitivity, and offered emotional support, positive feedback, and reassurance appeared to be important for parenting:

*"Nurses who were friendly and supportive toward their needs provided a conducive environment for maternal–new-born bonding."*[321]

*"Mothers and fathers emphasized the importance of reassurance from the nurse as a main contributing factor to their confidence in parenting, including normalizing the parent's emotional response, encouraging and praising the parent's efforts to care for their baby, and emphasizing the importance of the parent's contribution."* [308]

Also, proper information-provision and engaging parents in treatment discussions was reported to positively impact bonding as well as empowerment.[52,294,297,310,355,407] Parent participation in decision-making, in particular, was felt to be central to the parental identity.[291,305]

*"[There is] a strong need for the provision of accurate information regarding the infant's care and ongoing communication with professional staff in order to facilitate the formation of attachment."* [297]

*"Participants appreciated and valued their involvement, and decision making was perceived as an act of parental responsibility."* [305]

Negative communication effects reported included parents' feelings of incompetence and fear of caring for their infant.[226,308,380,381] In addition, negative communication experiences were said to cause emotional and physical distance between parents and infants, and induced feelings of not being a (good) parent at all[303,312,323,381]:

*"When parents felt that they were "in the way", excluded from decision making, excluded from their baby's care, or lacked access to information, they reported becoming depressed and seemed to have difficulties in bonding with and caring for their baby."* [381]

However, in quantitative studies, none of the reported interventions indeed showed significant effects on parenting. This applied to interventions across functions of communication, as well as to interventions focused on general communication strategies.

**3.4.5 Satisfaction.** *"Parents mentioned communication as the chief determinant of their satisfaction."*[401]

Finally, parents' satisfaction, both with overall care and with their relationships formed with individual health care professionals, was said to be affected by communication. Communication was reported to contribute positively to parents' overall perceptions of the quality of care provided by hospitals and NICU departments, as well as individual providers, notably also long-term.[294,297,299,300,314,324,392,401] Communication contributed to trusting relationships with care professionals.[294,298,300,321,346] Specifically, through experiencing emotional support and exchanging friendly conversations with nursing staff, receiving comprehensible information allowing parents to share their knowledge and involving them in medical rounds, parents' satisfaction with care was said to be enhanced:

*"Candid interactions with effective communication (frequent and informative exchanges, use of understandable terminology, willingness to answer questions); knowledgeable nurses who educate families; and emotional support (positivity, patience, respect, friendliness) contributed to a positive NICU experience and parent-nurse relationship."*[392]

Reported negative effects of communication included parents feeling humiliated, reprimanded, or offended and led to parents' overall dissatisfaction with care – also long-term, e.g., affecting parents' choice for a certain hospital during the next pregnancy.[302,305,313,352,380,382,383,394,401] In particular, poor relationships with nursing staff and a perceived lack of information-provision were mentioned as a cause for dissatisfaction.[302,314,383,392–394] Exclusion from care or decision-making were felt to hamper trusting relationships with staff:

*"When information shared was scarce, inaccurate, or "sugar coated," they were particularly dissatisfied."*[383]

*"If the parents were excluded or witnessed staff not responding to a baby quickly enough, it instilled a lack of confidence and mistrust in the staff."*[381]

Such tensions and conflict between parents and staff sometimes even resulted in requests to remove professionals from care, and occasionally also formal complaints:

*"When their concerns or suggestions about tests and treatments were dismissed, some parents activated the chain of command whereas others did not. However, those who did not activate the chain of command regretted not taking action."*[383]

Finally, quantitative records showed that satisfaction was increased significantly with interventions across all functions of communication. Also, overall communication interventions positively affected parents' satisfaction. When interventions focused on building and maintaining relationships between parents and staff, parents reported less decisional conflict.[397] Parents gained confidence in the healthcare team by being involved through family meetings and family-centred rounds.[385] Decision making aids caused conflicting results in terms of satisfaction: While one study reported increased satisfaction and less decisional conflict due to the implementation of a decision aid for the treatment of persistent ductus arteriosus[356], another study found a decrease in parents' satisfaction based on a decision-making tool within end-of-life care.[378]

# 4. DISCUSSION AND CONCLUSION

## 4.1 Discussion

This systematic review maps out five main effects (reported, observed, and established) of parent-provider interaction in the NICU on parents' (1) *coping* with the unexpected preterm birth of their infants, (2) *knowledge* of their infants' medical situation, (3) *participation* in communication and care in the NICU, (4) sense of empowerment and attachment in *parenting*, and (5) *satisfaction* with individual relationships with providers and overall care. These effects emerge across all functions of parent-provider communication in the NICU.[349]

Studies across medical domains report similar effects of language and interaction on patient-related outcomes. For instance, in oncology settings, several studies show effects of clinicians' empathy on patients' distress, trust, and satisfaction with care; effects of information-provision on both patients' recall of information and their participation in decision-making have been reported, too.[359–363,366] In general practice, the use of arguments to support treatment decisions has been shown to affect physician's credibility as well as their participatory style.[365] In the context of chronic care, communication has been argued to contribute to, e.g., patients' self-management.[409,410]

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Several of our findings, however, are specific to the NICU context. The reported effects of communication on parents' participation in infant care, their perceived parenting role, and the attachment to their infant are all characteristic to care that is provided to the parent-infant dyad in the NICU. Thereby, this study rather uniquely describes communication effects on patients' relatives and, more specifically, the impact of communication with providers on relatives' relationship to the patient. The literature on effects on relatives seems to be scant. Also, the reported longevity of effects may be particular to NICU communication. However, as communication studies often focus on short-term outcomes, this conclusion may be premature.

The present study thus confirms that in the NICU communication is one of the main predictors of quality of care as perceived by parents. While this finding is consistent with research in other domains[411–415], this is an important finding for conceptualizing and implementing Family Integrated Care within neonatal settings. Adequate communication gives parents a sense of being part of a team. Notably, not only relatively '*heavy*' or *structured* forms of communication (e.g., end-of-life decision-making or discharge planning) impact parents' perceptions. To parents, *day-to-day* communication with staff seems to be equally, if not more, important.[299,300,308,380] We believe this to be another particularity of interaction in NICU contexts. Our (qualitative) analyses reveal that 'chit-chat' concerning life outside of the NICU between parents and staff is valued highly and, at the same time, seemingly 'small' communication issues – misunderstandings, jokes, inadequate timing of information – carry a strong negative weight. Also, tone-of-voice matters a lot to parents in the interaction with medical professionals. These findings not only warrant further research, but also awareness from NICU staff. After all, bedside interaction is the most occurring form of communication in the NICU of all.

While the recent increase in studies on parent-provider communication shows that communication is no longer considered trivial in neonatal care, this review demonstrates that much more is needed to ensure that language and interaction become commonly recognized as vehicle to obtain optimal care outcomes. Parents still report suboptimal communication, rendering it important that communication training becomes part of all medical curricula. Within the Family Integrated Care model[55,66,86,226], not only staff education but also parents' inclusion in medical rounds is advocated to improve parent-staff exchanges. Nonetheless, our findings show that family-centred rounds are still often conceptualized as meetings in which parents merely meet with staff and ask questions. We believe that this communicative role is too passive. This may be addressed, e.g., by adding relational communication strategies to the Family Integrated Care model, as previously proposed by Benzies et al.[95,416] In order for parents to become active agents in their infants' care, it is crucial that a shift is established towards truly including parents in rounds as *equal* members of the care team, across all levels of NICU care.

Notably, this review shows that qualitative approaches are dominant in the study of parent-staff interaction and randomized controlled trials to test the efficacy of communication interventions are largely lacking. While our qualitative and quantitative findings all point in the same direction and, importantly, interviews, surveys, and observational studies can yield valuable insights into the experienced, self-reported, and observed effects of parent-provider interactions in neonatal care, obtaining reliable and reproducible data on communication effects remains crucial. While we recognize that it may be challenging to study communication *in vivo* using experimental studies, we strongly urge future studies to focus on testing causality between parent-provider interaction and outcomes, preferably in randomized controlled trials. Inspiration could be drawn from experimental research on communication in other medical domains.[359–363,365,366,417] Experiments should ideally focus on testing the effects of specific functions of communication. Also, potential adverse effects of communication should be more carefully explored.[378] Knowledge of the cause-effect relationships between communication and outcomes is essential when striving to develop evidence-based trainings for NICU staff.

The present study has several strengths. These include the comprehensive search strategy; the use of a quality assessment tool for mixed study designs; and the inclusion of communication scholars, medical doctors, and parent representatives in our research team. Yet, the results also need to be interpreted in light of some limitations. First, four articles could not be retrieved and had to be excluded. Second, the study purposefully focused on communication in a broad sense, rather than on the effectiveness of a single intervention, rendering a meta-analysis inappropriate. Third, the vast majority of included studies focus on birth mothers' perspectives rather than on their partners'. This is something that is commonly observed in the literature on parents' experiences of NICU care.[68] Yet, as a result, we could not make a distinction between the effects of communication by parenting roles. This needs to be systematically addressed in future studies, as the communicative interactions with staff may have an equally great yet different effect on partners.



### 4.2 Conclusion

Our results show that communication can tangibly improve parent-related outcomes in the NICU, both in the short- and the long-term: through providers' use of compassionate and supportive language; via accurate, accessible, timely information exchanges; by offering opportunities, structures, and tools for parents to engage in decision-making in their preferred role, and by empowering parents to assume their role as primary caregiver – both in the NICU and beyond. These findings confirm and elaborate the NICU Communication Framework.

### 4.3 Practice Implications

The NICU Communication Framework can be used as a starting point for improving parent-provider communication, through research and in practice. Our results show that hands-on communication interventions, such as written (supplementary) information or the adoption of family-centred rounds, can improve outcomes for NICU *parents* considerably. Moreover, there also seem to be indirect effects on preterm *infants*. The most evident example thereof is the reported effect of communication on parent-infant attachment. Positive effects of communication on *staff* work satisfaction can be assumed. Finally, by increasing parents' satisfaction with NICU staff members, neonatal wards and hospitals, and overall care, conflicts and (formal) complaints can potentially be reduced, ultimately also affecting the *health system* at large.

As such, we argue that it is time for communication to be seriously considered as a crucial determinant for parents' health and well-being, during and beyond infant admission to the NICU. NICU providers should particularly consider the impact of their day-to-day interactions with the family of ill infants, across all functions of communication. For, communication in the NICU seems to have the potential to harm as well as to heal parents.

**Table 1. Overview included records**

| Article reference | Country | NICU | | Sample | | Methods | | Included in |
|---|---|---|---|---|---|---|---|---|
| | | N | Levels | Family members | Staff members | Design | Type | |
| Abdel-Latif et al. (2015)[373] | Australia | 1 | 3 | 53 mothers<br>10 fathers | 1 doctor<br>7 nurses<br>1 pharmacist | mixed | RCT (cross-over, single-centre, non-blinded, randomized) and focus groups | both |
| Able-Boone et al. (1989)[288] | USA | 1 | 2 and 3 | 16 mothers<br>11 fathers | 15 doctors<br>15 nurses | qualitative | open-ended interviews (ethnographic) | meta-synthesis |
| Aija et al. (2019)[186] | Finland, Sweden, Norway, Estonia, Spain, Italy | 11 | 2 and 3 | 211 mothers<br>144 fathers | N/A | quantitative | multi-centred prospective surveys via text message | narrative synthesis |
| Alderson et al. (2006)[352] | UK | 4 | 3 | 80 mothers<br>16 fathers | 40 doctors | qualitative | semi-structured interviews and observations of informal contact between staff and parents (ethnographic) | meta-synthesis |
| AlFaleh et al. (2011)[356] | Saudi Arabia | 1 | 3 | 10 mothers | N/A | quantitative | surveys, pre-/post-test (pilot) | narrative synthesis |
| Arockiasamy et al. (2008)[289] | Canada | 1 | 3 | 16 fathers | N/A | qualitative | semi-structured interviews | meta-synthesis |
| Axelin et al. (2018)[226] | Finland | 1 | 3 | 15 mothers<br>7 fathers | 2 doctors | qualitative | interviews and video-recorded observations | meta-synthesis |
| Aydon et al. (2018)[290] | Australia | 1 | 3 | 20 mothers<br>20 fathers | N/A | qualitative | interviews | meta-synthesis |
| Baughcum et al. (2019)[374] | USA | 1 | 4 | 40 mothers<br>27 fathers | N/A | quantitative | surveys | narrative synthesis |

**Table 1. Overview included records (continued)**

| Article reference | Country | NICU | | Sample | | Methods | | Included in |
|---|---|---|---|---|---|---|---|---|
| | | N | Levels | Family members | Staff members | Design | Type | |
| Baughcum et al. (2017)[291] | USA | 1 | 4 | 42 mothers<br>28 fathers | N/A | qualitative | interviews and surveys | meta-synthesis |
| Berman et al. (2019)[292] | USA | N/A | 2 and 3 | 14 mothers<br>1 father | N/A | qualitative | interviews | meta-synthesis |
| Bramwell et al. (2005)[375] | UK | 34 | 3 | 59 mothers<br>24 fathers<br>3 grandparents | N/A | mixed | survey-based interviews, including open-ended and closed questions | meta-synthesis |
| Brinchmann et al. (2002)[293] | Norway | N/A | N/A | 19 mothers<br>16 fathers | N/A | qualitative | unstructured interviews | meta-synthesis |
| Broom et al. (2017)[294] | Australia | 1 | 3 | 4 mothers<br>1 grandparent | 8 nurses | qualitative | focus groups | meta-synthesis |
| Caeymaex et al. (2013)[376] | France | 4 | 3 | 48 mothers<br>30 fathers | N/A | mixed | semi-structured interviews and surveys | meta-synthesis |
| Cartwright et al. (2011)[377] | UK | 7 | 3 | 10 mothers<br>6 fathers | N/A | qualitative | semi-structured interviews | meta-synthesis |
| Clarke-Pounder et al. (2015)[378] | USA | 1 | 3 | 19 families† | N/A | quantitative | RCT (pre-/post intervention survey) and video-observations | narrative synthesis |
| Cox et al. (2001)[297] | UK | 1 | 3 | 241 mothers<br>10 other | N/A | mixed | (non-)participant observations, unstructured interviews, focus groups, and surveys | meta-synthesis |
| Enke et al. (2017)[269] | Germany | 66 | 3 | 1277 parents | N/A | quantitative | surveys | narrative synthesis |

**Table 1. Overview included records (continued)**

| Article reference | Country | NICU | | Sample | | Methods | | Included in |
|---|---|---|---|---|---|---|---|---|
| | | N | Levels | Family members | Staff members | Design | Type | |
| Epstein et al. (2015)[379] | USA | 1 | 3 | 15 parents | N/A | mixed | surveys | both |
| Falck et al. (2016)[298] | USA | 1 | 4 | 6 mothers | 1 doctor<br>5 nurses | qualitative | semi-structured interviews | meta-synthesis |
| Feeley et al. (2016)[52] | Finland, Canada | 2 | 3 | N/A | 37 nurses | qualitative | nurses' audio-recorded thoughts and perceptions | meta-synthesis |
| Fenwick et al. (2000)[299] | Australia | 2 | 2 and 3 | 31 families[†] | N/A | qualitative | unstructured interviews and audiotaped bedside interactions | meta-synthesis |
| Fenwick et al. (2001a)[300] | Australia | 2 | 2 and 3 | 28 mothers | 18 nurses<br>2 midwives (in training)[*] | qualitative | unstructured interviews and audiotaped bedside interactions | meta-synthesis |
| Fenwick et al. (2001b)[380] | Australia | 2 | 2 and 3 | 28 mothers | 20 nurses[*] | qualitative | unstructured interviews and audiotaped bedside interactions | meta-synthesis |
| Franck et al. (2017)[381] | UK | 7 | 3 | 33 mothers<br>7 fathers | N/A | qualitative | focus groups | meta-synthesis |
| Franck et al. (2012)[382] | UK | 4 | 3 | 169 parents | N/A | qualitative | Surveys with open-ended questions | meta-synthesis |
| Frize et al. (2013)[355] | Canada | 1 | 3 | 8 parents | 5 doctors | qualitative | surveys and observations (usability testing) | meta-synthesis |
| Gadepalli et al. (2017)[383] | worldwide | N/A | N/A | 96 mothers<br>14 fathers | N/A | mixed | surveys, including open-ended and closed questions | meta-synthesis |



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**Table 1. Overview included records (continued)**

| Article reference | Country | NICU | | Sample | | Methods | | Included in |
|---|---|---|---|---|---|---|---|---|
| | | N | Levels | Family members | Staff members | Design | Type | |
| Gallagher et al. (2018)[346] | UK | 1 | 3 | 12 mothers<br>6 fathers | N/A | qualitative | semi-structured interviews | meta-synthesis |
| Geetanjli et al. (2012)[302] | India | 2 | 3 | 16 parents | N/A | qualitative | interviews | meta-synthesis |
| Gonya et al. (2013)[384] | USA | 1 | 3 | 32 mothers | N/A | quantitative | surveys | narrative synthesis |
| Grzyb et al. (2014)[385] | Canada | 1 | 3 | 79 mothers<br>2 fathers | 68 doctors (in training)<br>28 nurses | quantitative | surveys | narrative synthesis |
| Guillaume et al. (2013)[303] | France | 3 | 3 | 30 mothers<br>30 fathers | N/A | qualitative | semi-directive interviews | meta-synthesis |
| Gustafson et al. (2016)[386] | USA | 1 | 3 | 78 mothers<br>54 fathers | N/A | quantitative | quasi-experiment | narrative synthesis |
| Harvey et al. (2013)[304] | UK | 1 | 3 | 13 mothers<br>5 fathers | N/A | qualitative | semi-structured interviews | meta-synthesis |
| Hasanpour et al. (2017)[387] | Iran | 3 | 3 | 203 parents | N/A | quantitative | surveys | narrative synthesis |
| Hendriks et al. (2017)[305] | Switzerland | 1 | 3 | 12 mothers<br>8 fathers | N/A | qualitative | semi-structured interviews | meta-synthesis |
| Hendson et al. (2015)[388] | Canada | 2 | 3 | N/A | 6 doctors<br>36 nurses<br>3 nurses (in training)<br>6 respiratory therapists<br>4 social workers<br>1 administrative staff<br>2 registered dieticians** | qualitative | focus groups | meta-synthesis |

**Table 1. Overview included records (continued)**

| Article reference | Country | NICU | | Sample | | Methods | | Included in |
|---|---|---|---|---|---|---|---|---|
| | | N | Levels | Family members | Staff members | Design | Type | |
| Ichijima et al. (2011)[307] | New Zealand, Japan | 2 | 2 and 3 | 61 mothers<br>60 fathers | N/A | mixed | surveys within semi-structured interviews | meta-synthesis |
| Ingram et al. (2017)[389] | UK | 4 | 2 | 37 parents | 5 doctors<br>18 nurses | qualitative | semi-structured interviews | meta-synthesis |
| Jones et al. (2015)[308] | Australia | 2 | 2 and 3 | 27 mothers<br>5 fathers | 12 nurses | qualitative | semi-structured interviews | meta-synthesis |
| Kavanaugh et al. (2010)[309] | USA | 3 | 3 | 40 mothers<br>14 fathers | 42 doctors<br>29 nurses | qualitative | semi-structured interviews | meta-synthesis |
| Kavanaugh et al. (2005)[310] | USA | 2 | 3 | 6 mothers<br>2 fathers | 6 doctors<br>2 nurses | qualitative | interviews | meta-synthesis |
| Kodjebacheva et al. (2017)[311] | USA | 1 | 3 | 6 mothers<br>2 fathers | 3 doctors<br>17 nurses | qualitative | focus groups | meta-synthesis |
| Lee et al. (2019)[390] | Korea | 1 | N/A | 54 parents | N/A | quantitative | surveys | narrative synthesis |
| Lemmen et al. (2013)[312] | Sweden | 3 | 3 and 4 | 12 families† | N/A | qualitative | semi-structured interviews | meta-synthesis |
| Lemmon et al. (2016)[313] | USA | 1 | 4 | 20 parents | N/A | qualitative | semi-structured interviews | meta-synthesis |

**Table 1. Overview included records (continued)**

| Article reference | Country | NICU | | Sample | | Methods | | Included in |
|---|---|---|---|---|---|---|---|---|
| | | N | Levels | Family members | Staff members | Design | Type | |
| Lemmon et al. (2018)[391] | USA | 1 | 3 | 30 parents | 30 health care professionals (including at least 1 neonatology attending, neonatology fellow, nurse practitioner, neonatal nurse, and social worker; paediatric neurology attending; paediatric palliative care attending; and paediatric chaplain) | mixed | semi-structured interviews, focus groups, and surveys | meta-synthesis |
| Martin et al. (2016)[392] | USA | 27 | 3 | 116 mothers<br>4 fathers | N/A | qualitative | surveys with open-ended questions | meta-synthesis |
| Modé et al. (2014)[314] | Sweden | 2 | 3 | 8 fathers | N/A | qualitative | semi-structured interviews | meta-synthesis |
| Moro et al. (2011)[393] | USA | 3 | 3 | 5 mothers | 4 doctors<br>4 nurses | qualitative | semi-structured interviews | meta-synthesis |
| Nicholas et al. (2014)[394] | Canada | 2 | 3 | N/A | 6 doctors<br>36 nurses<br>3 nurses (in training)<br>6 respiratory therapists<br>4 social workers<br>1 administrative staff<br>2 registered dieticians** | qualitative | focus groups | meta-synthesis |
| O'Brien et al. (2018)[55] | worldwide | 26 | 3 | 1443 mothers | N/A | quantitative | RCT (multicentre, cluster) | narrative synthesis |

**Table 1. Overview included records (continued)**

| Article reference | Country | NICU | | Sample | | Methods | | Included in |
|---|---|---|---|---|---|---|---|---|
| | | N | Levels | Family members | Staff members | Design | Type | |
| O'Brien et al. (2013)[56] | Canada | 1 | 3 | 42 mothers | N/A | mixed | prospective matched case-control study with cohort design (pilot) and semi-structured interviews | both |
| Palomaa et al. (2016)[395] | Finland | 7 | 2 and 3 | 106 mothers<br>34 fathers | N/A | qualitative | surveys with open-ended questions | meta-synthesis |
| Patriksson et al. (2019)[396] | Sweden | 3 | 2 and 3 | 10 parents | 10 health care professionals | qualitative | participant observations and interviews | meta-synthesis |
| Penticuff et al. (2005)[397] | USA | 2 | 3 | 122 mothers | N/A | quantitative | quasi-experiment | narrative synthesis |
| Phuma-Ngaiyaye et al. (2016)[321] | Malawi | 1 | 3 | 10 mothers | 5 nurses | qualitative | in-depth interviews | meta-synthesis |
| Pinch et al. (1993)[398] | USA | 1 | 3 | 28 mothers<br>18 fathers<br>1 grandparent | N/A | qualitative | interviews | meta-synthesis |
| Pinheiro et al. (2009)[399] | Brazil | 1 | N/A | 5 mothers<br>1 father | N/A | qualitative | semi-structured interviews | meta-synthesis |
| Raffray et al. (2014)[322] | Colombia | 1 | 2 and 3 | N/A | 3 doctors<br>11 nurses<br>1 speech therapist | qualitative | semi-structured interviews | meta-synthesis |
| Rodrigues et al. (2019)[323] | Brazil | 1 | 3 | N/A | 19 nurses | qualitative | semi-structured interviews | meta-synthesis |

**Table 1. Overview included records (continued)**

| Article reference | Country | NICU | | Sample | | Methods | | Included in |
|---|---|---|---|---|---|---|---|---|
| | | N | Levels | Family members | Staff members | Design | Type | |
| Russell et al. (2014)[324] | UK | 3 | 3 | 32 mothers<br>7 fathers | N/A | qualitative | interviews | meta-synthesis |
| Sabnis et al. (2018)[400] | USA | 1 | 4 | 64 parents | N/A | quantitative | surveys | narrative synthesis |
| Sankar et al. (2017)[401] | India | 1 | 3 | 100 parents | N/A | mixed | semi-structured surveys | both |
| Silva et al. (2010)[325] | Portugal | N/A | N/A | 14 mothers | 13 doctors<br>1 nurse | qualitative | semi-structured interviews | meta-synthesis |
| Smith et al. (2012)[402] | USA | 1 | 3 | 20 mothers<br>9 fathers | N/A | qualitative | semi-structured interviews | meta-synthesis |
| Trujillo et al. (2017)[403] | USA | 1 | N/A | N/A | N/A | quantitative | surveys | narrative synthesis |
| van de Vijver et al. (2015)[404] | UK | 1 | N/A | 105 parents | N/A | quantitative | quasi-experiment (pre-post intervention surveys) | narrative synthesis |
| van den Berg et al. (2011)[405] | Sweden | 1 | 3 | 223 parents | N/A | quantitative | surveys (pre-post design) | narrative synthesis |
| Verhagen et al. (2009)[406] | Netherlands | 10 | 3 | N/A | 147 doctors | qualitative | semi-structured interviews | meta-synthesis |
| Voos et al. (2011)[154] | USA | 1 | 3 | 28 parents | N/A | quantitative | surveys (pre-post design) | narrative synthesis |
| Weis et al. (2015)[327] | Denmark | 1 | 3 | 12 mothers<br>10 fathers | N/A | qualitative | semi-structured interviews | meta-synthesis |

**Table 1. Overview included records (continued)**

| Article reference | Country | NICU | | Sample | | Methods | | Included in |
|---|---|---|---|---|---|---|---|---|
| | | N | Levels | Family members | Staff members | Design | Type | |
| Weis et al. (2013)[407] | Denmark | 1 | 3 | 75 mothers<br>59 fathers | N/A | quantitative | RCT | narrative synthesis |
| Weiss et al. (2010)[408] | USA | 1 | 3 | 84 parents | N/A | quantitative | surveys (pre-post design) | narrative synthesis |
| Wigert et al. (2013)[347] | Sweden | 1 | 3 | 226 parents | N/A | mixed | surveys | meta-synthesis |
| **Total:** | | | | **6960 parents** (3499 mothers; 715 fathers; 15 other family members; 2731 unknown) | **693 staff members** (361 doctors (in training); 275 nurses (in training); 57 others) | | | |

* Represents the exact same participant pool. Participants have been counted once towards the total number of included parents and providers
** Represents the exact same participant pool. Participants have been counted once towards the total number of included parents and providers
† Assuming that only one family member participated, each family was counted once towards the total number of included parents

**Table 2. Quality assessment (QATSDD), each item received a score between 0-3**

| Records | | | QASTDD | | | | | | | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Reference | Characteristics | | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 | 11 | 12 | 13 | 14 | 15 | 16 | Scores | |
| | Type of study | Pilot study | Explicit theoretical framework AND/OR a comprehensive review of the literature | Statement of aims/objectives in main body of report | Clear description of research setting | Evidence of sample size considered in terms of analysis | Representative sample of target group of a reasonable size | Description of procedure for data collection | Rationale for choice of data collection tool(s) | Detailed recruitment data | Statistical assessment of reliability and validity of measurement tool(s) [Quantitative records] | Fit between research question and method of data collection [Quantitative records] | Fit between research question and format/method of data collection and/or content of data collection tool [Qualitative records] | Fit between research question and method of analysis | Good justification for analytic method selected | Assessment of reliability of analytic process [Qualitative records] | Evidence of user involvement in design [N/A for pilot studies] | Strengths and limitations critically discussed | Total score | Denominator (maximum score) |
| Abdel-Latif et al. (2015)[373] | mixed | no | 2 | 2 | 3 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 3 | 3 | 2 | 0 | 0 | 2 | **31** | 48 |
| Able-Boone et al. (1989)[288] | qualitative | no | 1.5 | 2.5 | 2.5 | 0.5 | 2.5 | 3 | 3 | 2 | | | 3 | 3 | 2.5 | 2.5 | 0 | 2 | **30.5** | 42 |
| Aija et al. (2019)[186] | quantitative | no | 2 | 2.5 | 3 | 1 | 2.5 | 2.5 | 2 | 2 | 0 | 2 | | 2.5 | 3 | | 0 | 1.5 | **26.5** | 42 |
| Alderson et al. (2006)[352] | qualitative | no | 1 | 2 | 3 | 1 | 2 | 2 | 2 | 2 | | | 3 | 2 | 1 | 0 | 0 | 1 | **22** | 42 |
| AlFaleh et al. (2011)[356] | quantitative | yes | 0.5 | 2.5 | 1.5 | 0 | 0.5 | 1 | 1 | 0.5 | 0.5 | 2 | | 1.5 | 1 | | N/A | 1 | **13.5** | 39 |
| Arockiasamy et al. (2008)[289] | qualitative | no | 2 | 2 | 3 | 3 | 3 | 2.5 | 1 | 0.5 | | | 2.5 | 3 | 2.5 | 3 | 0 | 1.5 | **29.5** | 42 |

**Table 2. Quality assessment (QATSDD), each item received a score between 0-3 (continued)**

| Records | | | QASTDD | | | | | | | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Reference | Characteristics | | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 | 11 | 12 | 13 | 14 | 15 | 16 | Scores | |
| Axelin et al. (2018)[226] | qualitative | no | 2 | 2.5 | 3 | 1.5 | 3 | 2 | 2.5 | 2.5 | | | 2.5 | 3 | 2.5 | 2 | 0 | 2.5 | **31.5** | 42 |
| Aydon et al. (2018)[290] | qualitative | no | 3 | 2 | 3 | 2.5 | 2 | 3 | 2.5 | 3 | | | 3 | 2.5 | 3 | 3 | 0 | 2.5 | **35** | 42 |
| Baughcum et al. (2019)[374] | quantitative | no | 3 | 2.5 | 3 | 1 | 2 | 3 | 2.5 | 3 | 2.5 | 3 | | 2 | 1.5 | | 0 | 3 | **32** | 42 |
| Baughcum et al. (2017)[291] | qualitative | yes | 2 | 3 | 3 | 3 | 3 | 3 | 2 | 3 | | | 2 | 3 | 2 | 3 | N/A | 2 | **34** | 39 |
| Berman et al. (2019)[292] | qualitative | no | 1 | 1 | 1.5 | 3 | 2 | 1.5 | 1.5 | 0.5 | | | 2 | 3 | 2 | 3 | 0 | 1.5 | **23.5** | 42 |
| Bramwell et al. (2005)[375] | mixed | no | 1 | 1.5 | 3 | 0 | 1 | 1 | 0 | 1 | 0 | 1 | 0 | 1 | 0 | 0 | 0 | 1 | **11.5** | 48 |
| Brinchmann et al. (2002)[293] | qualitative | no | 1 | 1 | 2 | 1 | 2 | 2 | 0 | 2 | | | 2 | 2 | 1 | 1 | 0 | 2 | **19** | 42 |
| Broom et al. (2017)[294] | qualitative | no | 2.5 | 2 | 3 | 0 | 1.5 | 2 | 1.5 | 1.5 | | | 2 | 2 | 1 | 1.5 | 0 | 2 | **22.5** | 42 |
| Caeymaex et al. (2013)[376] | mixed | no | 2 | 3 | 2 | 2 | 3 | 3 | 3 | 3 | 1 | 3 | 2 | 2 | 2 | 3 | 0 | 2 | **36** | 48 |
| Cartwright et al. (2011)[377] | qualitative | no | 2 | 3 | 2 | 0 | 1.5 | 3 | 1.5 | 1.5 | | | 2 | 2 | 1.5 | 1.5 | 0 | 2.5 | **24** | 42 |
| Clarke-Pounder et al. (2015)[378] | quantitative | yes | 1 | 2 | 1.5 | 0 | 2 | 2 | 1 | 2 | 0.5 | 1.5 | | 1.5 | 1 | | N/A | 0 | **16** | 39 |
| Cox et al. (2001)[297] | mixed | no | 3 | 2.5 | 2 | 2 | 2.5 | 3 | 3 | 1.5 | 2 | 2 | 3 | 3 | 3 | 3 | 1 | 0 | **36.5** | 48 |
| Enke et al. (2017)[269] | quantitative | no | 2.5 | 3 | 2 | 2 | 2 | 2.5 | 1 | 2.5 | 2.5 | 2.5 | | 2.5 | 3 | | | 2.5 | **30.5** | 42 |
| Epstein et al. (2015)[379] | mixed | yes | 2 | 2.5 | 3 | 0.5 | 1 | 2 | 1.5 | 1.5 | 1 | 1 | 1.5 | 1.5 | 1.5 | 0 | N/A | 2 | **22.5** | 45 |
| Falck et al. (2016)[298] | qualitative | no | 2.5 | 2 | 2.5 | 2.5 | 2.5 | 2.5 | 2 | 2 | | | 2.5 | 2.5 | 2 | 2.5 | 0 | 2 | **30** | 42 |
| Feeley et al. (2016)[52] | qualitative | no | 2.5 | 1.5 | 3 | 3 | 2 | 2.5 | 2.5 | 2 | | | 2.5 | 2.5 | 2 | 2.5 | 0 | 2 | **30.5** | 42 |
| Fenwick et al. (2000)[299] | qualitative | no | 2.5 | 2 | 2.5 | 0 | 1.5 | 2 | 2.5 | 1.5 | | | 2.5 | 2 | 2.5 | 2 | 0 | 0 | **23.5** | 42 |
| Fenwick et al. (2001a)[300] | qualitative | no | 2.5 | 2.5 | 2.5 | 0 | 2.5 | 2.5 | 2.5 | 1 | | | 3 | 2.5 | 2.5 | 0 | 0 | 0.5 | **24.5** | 42 |
| Fenwick et al. (2001b)[380] | qualitative | no | 2 | 3 | 3 | 0 | 2 | 3 | 2 | 1 | | | 3 | 3 | 3 | 3 | 0 | 0 | **28** | 42 |
| Franck et al. (2017)[381] | qualitative | no | 2 | 2.5 | 2.5 | 0 | 2.5 | 2.5 | 3 | 2 | | | 2.5 | 2.5 | 2 | 1.5 | 0 | 2 | **27.5** | 42 |
| Franck et al. (2012)[382] | qualitative | no | 3 | 3 | 2 | 0 | 0.5 | 2 | 1.5 | 1 | | | 2.5 | 3 | 3 | 3 | 0 | 1 | **25.5** | 42 |
| Frize et al. (2013)[355] | qualitative | no | 2 | 3 | 2 | 0 | 0 | 1 | 0 | 0 | | | 1 | 0 | 0 | 0 | 0 | 0 | **9** | 42 |

**Table 2. Quality assessment (QATSDD), each item received a score between 0-3 (continued)**

| Records | | | QASTDD | | | | | | | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Reference | Characteristics | | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 | 11 | 12 | 13 | 14 | 15 | 16 | Scores | |
| Gadepalli et al. (2017)[383] | mixed | no | 2.5 | 2.5 | 1 | 0 | 1.5 | 1.5 | 1 | 2 | 3 | 3 | 1.5 | 3 | 1.5 | 1.5 | 2 | 2 | **29.5** | 48 |
| Gallagher et al. (2018)[346] | qualitative | no | 1.5 | 2 | 2 | 2 | 2 | 2 | 0.5 | 2 | | | 2.5 | 2.5 | 0.5 | 1.5 | 0 | 1.5 | **22.5** | 42 |
| Geetanjli et al. (2012)[302] | qualitative | no | 1 | 1 | 1 | 0 | 1 | 1 | 1 | 1 | | | 2 | 2 | 0 | 0 | 0 | 1 | **12** | 42 |
| Gonya et al. (2013)[384] | quantitative | no | 1 | 2.5 | 3 | 0 | 1.5 | 1 | 1.5 | 1.5 | 0 | 1 | | 1.5 | 1.5 | | 0 | 1.5 | **17.5** | 42 |
| Grzyb et al. (2014)[385] | quantitative | no | 1 | 3 | 3 | 0 | 1.5 | 1.5 | 1.5 | 3 | 0 | 1.5 | | 2 | 1.5 | | 0 | 2.5 | **22** | 42 |
| Guillaume et al. (2013)[303] | qualitative | no | 3 | 3 | 2 | 3 | 2 | 2 | 2 | 3 | | | 3 | 3 | 1 | 1 | 2 | 2 | **32** | 42 |
| Gustafson et al. (2016)[386] | quantitative | no | 2 | 3 | 2 | 3 | 2 | 3 | 2 | 3 | 3 | 3 | | 2 | 2 | | 0 | 2 | **32** | 42 |
| Harvey et al. (2013)[304] | qualitative | no | 2 | 2 | 2 | 2.5 | 2 | 2 | 2.5 | 1.5 | | | 2 | 2 | 1.5 | 1.5 | 0 | 1.5 | **25** | 42 |
| Hasanpour et al. (2017)[387] | quantitative | no | 2 | 3 | 1 | 2 | 0 | 2 | 2 | 2 | 2 | 3 | | 1 | 1 | | 2 | 1 | **24** | 42 |
| Hendriks et al. (2017)[305] | qualitative | no | 1 | 1 | 2 | 1 | 2 | 1 | 1 | 1 | | | 2 | 1 | 1 | 0 | 0 | 1 | **15** | 42 |
| Hendson et al. (2015)[388] | qualitative | no | 2.5 | 3 | 2.5 | 2 | 2 | 1.5 | 2.5 | 0.5 | | | 3 | 3 | 2.5 | 2.5 | 0 | 2 | **29.5** | 42 |
| Ichijima et al. (2011)[307] | mixed | no | 2 | 2 | 3 | 0 | 3 | 2 | 2 | 1 | 2 | 3 | 3 | 3 | 2 | 0 | 0 | 1 | **29** | 48 |
| Ingram et al. (2017)[389] | qualitative | no | 2.5 | 1 | 2 | 0 | 2.5 | 2 | 1 | 1 | | | 2 | 2 | 1 | 2 | 1.5 | 2 | **22.5** | 42 |
| Jones et al. (2015)[308] | qualitative | no | 3 | 2 | 3 | 3 | 1.5 | 1.5 | 0.5 | 1 | | | 2.5 | 2.5 | 2 | 2 | 0 | 2.5 | **27** | 42 |
| Kavanaugh et al. (2010)[309] | qualitative | no | 2 | 2 | 1 | 0 | 2 | 3 | 2 | 1 | | | 3 | 3 | 1 | 3 | 0 | 1 | **24** | 42 |
| Kavanaugh et al. (2005)[310] | qualitative | yes | 3 | 2 | 1.5 | 0 | 1 | 3 | 2.5 | 1.5 | | | 2 | 2 | 1.5 | 0 | N/A | 1 | **21** | 39 |
| Kodjebacheva et al. (2017)[311] | qualitative | no | 2.5 | 3 | 3 | 0 | 1 | 2 | 0.5 | 0.5 | | | 2 | 2 | 1 | 1.5 | 0 | 2 | **21** | 42 |
| Lee et al. (2019)[390] | quantitative | no | 3 | 3 | 2 | 3 | 3 | 3 | 3 | 1 | 3 | 3 | | 2 | 3 | | 0 | 1 | **33** | 42 |
| Lemmen et al. (2013)[312] | qualitative | no | 3 | 3 | 3 | 0 | 2 | 2.5 | 2 | 1.5 | | | 3 | 3 | 2.5 | 3 | 0 | 2.5 | **31** | 42 |
| Lemmon et al. (2016)[313] | qualitative | no | 1 | 2 | 2 | 3 | 3 | 1 | 2 | 1 | | | 2 | 2 | 1 | 2 | | 3 | **25** | 42 |
| Lemmon et al. (2018)[391] | mixed | no | 1 | 3 | 3 | 3 | 3 | 2 | 3 | 2 | 0 | 1 | 3 | 3 | 3 | 3 | 3 | 2 | **38** | 48 |

**Table 2. Quality assessment (QATSDD), each item received a score between 0-3 (continued)**

| Records | | | QASTDD | | | | | | | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Reference | Characteristics | | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 | 11 | 12 | 13 | 14 | 15 | 16 | Scores | |
| Martin et al. (2016)[392] | qualitative | no | 2 | 2 | 2 | 0 | 0 | 1 | 1 | 2 | | | 1 | 2 | 2 | 0 | 0 | 1 | **16** | 42 |
| Modé et al. (2014)[314] | qualitative | no | 2 | 2 | 2 | 0 | 1.5 | 2 | 2 | 1 | | | 2 | 2.5 | 1.5 | 1.5 | 0 | 1.5 | **21.5** | 42 |
| Moro et al. (2011)[393] | qualitative | no | 2 | 2 | 1 | 0 | 1 | 2 | 3 | 1 | | | 2 | 2 | 2 | 2 | 0 | 2 | **22** | 42 |
| Nicholas et al. (2014)[394] | qualitative | no | 3 | 2.5 | 3 | 2.5 | 2.5 | 3 | 3 | 2.5 | | | 2.5 | 2.5 | 2.5 | 1.5 | 0 | 2 | **33** | 42 |
| O'Brien et al. (2013)[56] | mixed | yes | 3 | 3 | 1 | 1 | 2 | 3 | 2 | 3 | 2 | 3 | 2 | 2 | 2 | 0 | N/A | 3 | **32** | 39 |
| O'Brien et al. (2018)[55] | quantitative | no | 2.5 | 1.5 | 2 | 3 | 2 | 2 | 2.5 | 3 | 1 | 2.5 | | 2.5 | 1.5 | | 0 | 2.5 | **28.5** | 42 |
| Palomaa et al. (2016)[395] | qualitative | no | 3 | 3 | 3 | 2 | 2 | 2.5 | 2 | 2 | | | 2 | 2.5 | 2.5 | 2.5 | 1 | 2.5 | **32.5** | 42 |
| Patriksson et al. (2019)[396] | qualitative | no | 2 | 1.5 | 3 | 0 | 0.5 | 1 | 2 | 0.5 | | | 2 | 2 | 1.5 | 0 | 0 | 1.5 | **17.5** | 42 |
| Penticuff et al. (2005)[397] | quantitative | no | 2 | 2 | 2 | 0.5 | 2 | 3 | 2 | 1 | 2 | 1.5 | | 2 | 1.5 | | 0 | 0 | **21.5** | 42 |
| Phuma-Ngaiyaye et al. (2016)[321] | qualitative | no | 2.5 | 2 | 2 | 2.5 | 1 | 2 | 0.5 | 0.5 | | | 2 | 2 | 1.5 | 1.5 | 0 | 2 | **22** | 42 |
| Pinch et al. (1993)[398] | qualitative | no | 2 | 2 | 2 | 3 | 1 | 2 | 2.5 | 2 | | | 2.5 | 2.5 | 2 | 3 | 1 | 0.5 | **28** | 42 |
| Pinheiro et al. (2009)[399] | qualitative | no | 1 | 3 | 3 | 2 | 1 | 3 | 1 | 1 | | | 2 | 2 | 2 | 0 | 0 | 0 | **21** | 42 |
| Raffray et al. (2014)[322] | qualitative | no | 2.5 | 2.5 | 3 | 3 | 2 | 2.5 | 1 | 1.5 | | | 2.5 | 2 | 1.5 | 3 | 0 | 2 | **29** | 42 |
| Rodrigues et al. (2019)[323] | qualitative | no | 2 | 2 | 3 | 0 | 2 | 2.5 | 0.5 | 2 | | | 2 | 2.5 | 1.5 | 1 | 0 | 1.5 | **22.5** | 42 |
| Russell et al. (2014)[324] | qualitative | no | 2 | 1.5 | 2 | 2.5 | 2 | 2.5 | 2 | 2 | | | 2 | 2.5 | 2.5 | 0 | 0 | 3 | **26.5** | 42 |
| Sabnis et al. (2018)[400] | quantitative | no | 1 | 2 | 3 | 0 | 0 | 2 | 2 | 1 | 1 | 1 | | 2 | 2 | | 0 | 2 | **19** | 42 |
| Sankar et al. (2017)[401] | mixed | no | 2 | 2.5 | 1.5 | 0 | 1.5 | 2 | 0.5 | 0.5 | 0 | 1.5 | 1 | 1.5 | 0 | 0 | 0 | 1.5 | **16** | 48 |
| Silva et al. (2010)[325] | qualitative | no | 1 | 3 | 2 | 0.5 | 1 | 1.5 | 0.5 | 1 | | | 1.5 | 1.5 | 0.5 | 0 | 0 | 1.5 | **15.5** | 42 |
| Smith et al. (2012)[402] | qualitative | no | 1.5 | 2 | 2.5 | 3 | 2 | 2 | 2.5 | 1.5 | | | 2.5 | 2.5 | 2 | 2.5 | 0.5 | 2 | **29** | 42 |
| Trujillo et al. (2017)[403] | quantitative | no | 2 | 2 | 2 | 0 | 0 | 2 | 0 | 0 | 0 | 1 | | 2 | 0 | | 0 | 0 | **11** | 42 |
| van de Vijver et al. (2015)[404] | quantitative | no | 1.5 | 2.5 | 1 | 0 | 0.5 | 1.5 | 0 | 0 | 0 | 1.5 | | 1 | 0 | | 0 | 1 | **10.5** | 42 |

**Table 2. Quality assessment (QATSDD), each item received a score between 0-3 (continued)**

| Records | | | QASTDD | | | | | | | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Reference | Characteristics | | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 | 11 | 12 | 13 | 14 | 15 | 16 | Scores | |
| van den Berg et al. (2011)[405] | quantitative | no | 2 | 2 | 3 | 0 | 1.5 | 2.5 | 1.5 | 2 | 0 | 2 | | 1.5 | 1 | | 0 | 2 | **21** | 42 |
| Verhagen et al. (2009)[406] | qualitative | no | 1,5 | 2,5 | 2,5 | 0 | 1 | 1.5 | 1 | 1.5 | | | 2 | 2 | 1 | 1 | 0 | 2,5 | **14** | 42 |
| Voos et al. (2011)[154] | quantitative | no | 2.5 | 3 | 1 | 0 | 0.5 | 2.5 | 1.5 | 1.5 | 0 | 2.5 | | 2 | 1.5 | | 0 | 1.5 | **20** | 42 |
| Weis et al. (2015)[327] | qualitative | no | 3 | 3 | 3 | 3 | 3 | 3 | 2 | 2 | | | 3 | 3 | 2 | 3 | 0 | 2 | **35** | 42 |
| Weis et al. (2013)[407] | quantitative | no | 2 | 3 | 3 | 3 | 3 | 3 | 2 | 3 | 2 | 3 | | 3 | 2 | | 0 | 3 | **35** | 42 |
| Weiss et al. (2010)[408] | quantitative | no | 2 | 2 | 2 | 1 | 2 | 2 | 2 | 1 | 1 | 2 | | 2 | 1 | | 3 | 2 | **25** | 42 |
| Wigert et al. (2013)[347] | mixed | no | 3 | 3 | 3 | 3 | 2 | 2 | 1.5 | 2.5 | 1 | 1.5 | 2 | 2 | 1.5 | 0 | 1 | 2 | **31** | 48 |

**Table 3. Quantitative findings as reported in included records.**

| Functions | Interventions/exposures | Specific intervention/ exposure | Article reference | N = Parents *Intervention* | Gestational Age *Intervention* | N = Parents *Control* | Gestational Age *Control* | Outcomes** | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | | | | Coping | Knowledge | Participation | Parenting | Satisfaction |
| Building/maintaining relationships | | | | | | | | | | | | |
| | **1.** Inclusive communication | | | | | | | | | | | |
| | | **Care Planning Meetings** | Penticuff et al. (2005)[397] | 60 | unknown | 62 | unknown | The intervention group had fewer unrealistic concerns (p = .018) | The intervention group had less uncertainty about infant medical conditions (p = .003) | The intervention group reported more shared decision making with professionals than did the control group (p = .010) | | The intervention group was more satisfied with the amount of decision input they had (p = .058)<br><br>The intervention group had less decision conflict (p ≤ .001) and was more satisfied with the process by which medical decisions were made (p = .012)<br><br>There were no statistically significant differences between control and intervention groups in satisfaction with their infants' care, satisfaction with relationships with NICU physicians and nurses, and satisfaction with the decisions made for infant treatment |
| | | **Facilitating open parent-staff interaction** | Weiss et al. (2010)[408] | 50 | 32 (4.4) | 34 | 32 (9.0) | | Understanding of health care professionals' communication (68% vs 60%, p = 0.52) | More communication with health care professionals before birth (97% vs 76%, p < 0.01) and during their baby's admission (100% vs 83%, p = 0.01).<br><br>Reciprocity (ability to ask questions): 97% vs 90% (p = 0.24) | | |

**Table 3. Quantitative findings as reported in included records. (continued)**

| | | | | | | Outcomes** | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | | Coping | Knowledge | Participation | Parenting | Satisfaction |
| **2.** Supportive communication | | | | | | | | | | |
| **Empathy in communication** | Enke et al. (2017)[269] | 1277 | 29 | N/A | N/A | PSS-NICU communication stress (b = -0.130, p <0.001) | | | Associations between parental role stress and measures of perceived communication were trivial and could not be significantly distinguished from zero. | |
| Exchanging information | | | | | | | | | | |
| **1.** Information accessibility | | | | | | | | | | |
| **Video conferencing** | Epstein et al. (2015)[379] | 25 | 31 (4) | 25† | 31 (4) | Degree of parent worry about problems (p > 0.05) | Parents' understanding of their baby's care and outcomes in the NICU: Broad views of having infant in neonatal intensive care unit (p = 0.001), specific parent experiences in last week (p = 0.04), presence of problems common to premature infants (p > 0.05) | | | |
| **Baby diary** | Van de Vijver et al. (2015)[404] | 44 | N/A | 44† | N/A | | Understand why baby is admitted (100% vs. 100%). Updated by doctor < 24h of admission (100% vs 86%), updated throughout stay (93% vs 76%) | Questions and concerns addressed (93% vs 91%).<br>Feeling of being involved in baby's care (100% vs 88%) | | 100% found the diary good practice<br>100% liked reading the diary |

**Table 3. Quantitative findings as reported in included records. (continued)**

| | | | | | | Outcomes** | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | | Coping | Knowledge | Participation | Parenting | Satisfaction |
| **2.** Enabling parents to share information | | | | | | | | | | |
| **Family meetings** | Sabnis et al. (2018)[400] | 34 | 34.2 (95% CI 33.6–34.8) | 26 | 34.5 (95% CI: 34.0–35.1) | | | Proportion of parents having a timely family meeting (31.3% vs 10.5%, $p < 0.001$) | | Families' satisfaction with clinician communication ($p > 0.05$) |
| **Interdisciplinary family conferences** | Trujillo et al. (2017)[403] | unknown | < 32 | N/A | N/A | | IFCs within the first 14 days of admission increased family's understanding of their infant's plan of care | | | IFCs within the first 14 days of admission improved parents' satisfaction with the NICU team |
| (Sharing) decision making | | | | | | | | | | |
| **1.** Structures to facilitate decision-making | | | | | | | | | | |
| **Family-centred rounds** | Abdel-Latif et al. (2015)*[373] | 63 | 31 | 63† | 31 | PSS-NICU total score (3.439 (3.268 to 3.609) vs 3.471 (3.301 to 3.642), $p = 0.62$) | Received adequate information about baby's condition and management (4.321 (4.092 to 4.551) vs 3.947 (3.712 to 4.182), $p = 0.03$)<br><br>Things were explained thoroughly using easy to understand language (4.325 (4.114 to 4.537) vs 4.230 (4.013 to 4.446), $p = 0.49$)<br><br>Information received has been appropriate and timely (4.057 (3.845 to 4.270) vs 4.357 (4.119 to 4.595) $p = 0.13$) | Able to communicate effectively with baby's healthcare team ($p = 0.05$)<br><br>Collaborated with baby's healthcare team in the planning of care for baby ($p = 0.02$)<br><br>Able to ask the healthcare team questions about baby's care ($p = 0.004$) | Parental role stress (PSS-NICU, 3.477 (3.262 to 3.692) vs 3.638 (3.421 to 3.856) $p = 0.09$) | |

**Table 3. Quantitative findings as reported in included records. (continued)**

| | | | | | | Outcomes** | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | | Coping | Knowledge | Participation | Parenting | Satisfaction |
| **Medical rounds** | Aija et al. (2019)[186] | 241 | < 35 | N/A | N/A | | | When the unit's policy was to routinely invite parents to medical rounds, the mothers were more likely to be present (p = 0.036; fathers (p = 0.873) | | |
| **Family-centred rounds** | Grzyb et al. (2014)[385] | 81 | 34.1 (5.9) to 35.3 (5.0) | N/A | N/A | Attending rounds helped parents to be less worried about their child (84%).<br><br>34% found it upsetting when there was uncertainty expressed about the care or condition of child during rounds | The discussion during rounds was more confusing than helpful (8%)<br><br>Would've missed important information if not present (68%)<br><br>Too many medical terms were used (17%) | Felt comfortable asking questions during rounds (83%)<br><br>Parent being present for rounds improved the care of child (44%) | | Liked being present during rounds (98%)<br><br>Being present during rounds gave more confidence in the health-care team (88%)<br><br>Felt ignored during rounds (11%) |
| **Multidisciplinary rounds** | Gustafson et al. (2016)[386] | 86 | 38 (IQR: 24, 41) | 46 | 38 (IQR: 25, 41) | Overall PSS-NICU score -0.12 +0 0.10, p = .25 | | | PSS-NICU role alteration -0.26 +/- 0.14, p = .06 | |
| **Family-centred rounds** | Voos et al. (2011)[154] | 16 | N/A | 12 | NA | No difference in PSS-NICU score (3.35 vs 3.12, p = NS) | Being kept informed as to changes in the infant's condition, meeting with physicians, and information about long-term expectations: 5.5 vs 4.4 (p < 0.01) | 75% attended FCR multiple times per week. | | Overall Neonatal Instrument of Parent Satisfaction: 5.47 vs 5.26, p = NS |

**Table 3. Quantitative findings as reported in included records. (continued)**

| | | | | | | Outcomes** | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | | Coping | Knowledge | Participation | Parenting | Satisfaction |
| **2.** Decision aids | | | | | | | | | | |
| **Decision aid for patent ductus arteriosus** | AlFaleh et al. (2011)[356] | 10 | unknown | 10 | unknown | | Increase in knowledge, $p < 0.0001$<br><br>Increase in understandability, $p < 0.0001$ | | | Increase in satisfaction, $p < 0.0001$<br><br>Decisional conflict (0.6 (1.4), vs 25.4 (7.5), $p < 0.00001$) |
| **Decision-making tool** | Clarke-Pounder et al. (2015)[*378] | | 37 | | 30.5 | STAI - 0.33 (95% CI - 6.34–5.67) | For the question, "I needed to know what treatment my baby was receiving," the intervention group was less satisfied compared to the controls ($p = 0.013$) | The duration and the relative contribution of parents to rounds were not significantly different between groups | | Decrease of satisfaction among the intervention group compared to the control group across, although not statistically significant |
| Enabling self-management | | | | | | | | | | |
| **1.** Discharge planning | | | | | | | | | | |
| **Transport preparation** | Van den Berg et al. (2011)[405] | 180 | unknown | 43 | unknown | Families who had been in contact at least once with the staff at the community hospital (CH) rated their feeling of being prepared before the transition significantly higher ($p < 0.001$) than families who had no contact with the CH prior the transition | | | | Families who had at least one prior contact with the CH rated their reception at the CH significantly higher ($p < 0.001$) than those who had no prior contact |

**Table 3. Quantitative findings as reported in included records. (continued)**

| | | | | | | Outcomes** | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | | Coping | Knowledge | Participation | Parenting | Satisfaction |
| Communication across functions | | | | | | | | | | |
| **1.** Family-integrated care (FIC) | | | | | | | | | | |
| **Family-integrated care** | O'Brien et al. (2013)[56] | 42 | < 35 | 14 | < 35 | Mean parental stress scores at discharge had decreased among FIC mothers ($p < 0.01$). No significant change was noted in the control group ($p > 0.05$). | | | | |
| **Family-integrated care** | O'Brien et al. (2018)[*55] | 738 | unknown | 705 | unknown | Mean stress and anxiety scores for FIC parents were significantly lower than those for parents in the standard care group, as measured during a 21-day window any time during hospitalization. (stress $p < 0.00043$; anxiety $p = 0.0045$) | | | | |

**Table 3. Quantitative findings as reported in included records. (continued)**

| | | | | | | | Outcomes** | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | | | Coping | Knowledge | Participation | Parenting | Satisfaction |
| **2.** Guided Family Centred Care | | | | | | | | | | | |
| | **Guided Family Centred Care** | Weis et al. (2013)*[407] | 74 | 29.2 | 60 | 30.1 | PSS-NICU: total score; -0.14 (95% CI: -0.40; 0.12, p = 0.28)<br><br>Sights and sounds: -0.28 (95% CI -0.59; 0.03, p = 0.07), Infant behaviour: -0.22 (-0.49; 0.06, p = 0.12) | | Intervention group 70% ≥ 3 meetings, 18% 2 meetings, 11% had 1 meeting vs. control group: 50% ≥ 3 unscheduled meetings | PSS-NICU parental role: +0.07 (95% CI -0.28; 0.43, p = 0.69) | |
| **3.** End of life communication | | | | | | | | | | | |
| | **Quality of communication** | Baughcum et al. (2019)[374] | 67 | 32.8 (6.5) | N/A | N/A | High quality of communication during EOL correlates positively with emotional needs that are met well (mothers +0.70 (p < 0.0001), fathers +0.64 (p < 0.0001) | High quality of communication during EOL correlates positively with information/ knowledge that are met well (mothers +0.76 (p < 0.0001), fathers +0.82 (p < 0.0001) | High quality of communication during EOL correlates positively with the feeling of family inclusion (mothers +0.87 (p < 0.0001), fathers +0.86 (p < 0.0001) | | High quality of communication during EOL correlates positively with needs that are met well (mothers +0.79 (p < 0.0001), fathers +0.69 (p < 0.0001)<br><br>High quality of communication during EOL correlates positively with overall satisfaction with care scores (mothers +0.94 (p < 0.0001), fathers +0.91 (p < 0.0001) |
| **3.** General | | | | | | | | | | | |
| | **Parent-nurse interaction** | Gonya et al. (2013)[384] | 32 | All < 27<br><br>72% ≤ 25 | N/A | N/A | | | Skin-to-skin contact increases with higher communication scores (p = 0.175)<br><br>Maternal presence increases with higher communication scores (p = 0.091) | | |

**Table 3. Quantitative findings as reported in included records. (continued)**

| | | | | | | | Outcomes** | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | | | Coping | Knowledge | Participation | Parenting | Satisfaction |
| | **Parent-staff communication** | Hasanpour et al. (2017)[387] | 203 | unknown | N/A | N/A | PSS-NICU stress (r = −0.144, p = 0.041) | | | | |
| | **Nurses communication ability and communication type** | Lee et al. (2019)[390] | 54 | N=14 < 37<br><br>N=40 > 37 | N/A | N/A | Correlations between communication ability and PSS-NICU total score (-23, p = 0.095)<br><br>Correlations between communication style and PSS-NICU total score (informative: -.10, p = .456; friendly: -.23, p = .102; authoritative: .11, p = .424) | | | Correlations between communication ability and parental role stress (-.14, p = .326)<br><br>Correlations between communication style and parental role stress (informative: --.05, p = .729; friendly: -.14, p = .312; authoritative: .15, p = .280) | |
| | **Traditional NICU care** | Sankar et al. (2017)[401] | 100 | N= 67 > 34 weeks<br><br>N = 33 < 34 | N/A | N/A | Nearly one-third of the parents felt that doctors took very little time to answer their queries, and this increased their apprehension | Almost half of the parents stated that they felt the information provided to them by the caregivers was inadequate | | | Parents expressed fair satisfaction with regard to the emotional support and encouragement received<br><br>A large fraction of the parents mentioned communication as the chief determinant of their satisfaction. A little over half of the parents expressed their dissatisfaction with the communication with caregivers |

*RCT, **outcomes are noted first for intervention and then for control, † Control group and intervention group are the same participants. Abbreviations used, not explained in table: b (beta), CI (confidence interval), r (correlation coefficient), IQR (interquartile range,) NS (non-significant), PSS (Parental Stress Scale), STAI (State-Trait Anxiety Inventory)

# Part III

# An innovative neonatal FICare model

Chapter 9

# Association of a Zero-Separation Neonatal Care Model With Stress in Mothers of Preterm Infants.

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# ABSTRACT


**Importance**

Active participation in care by parents and zero-separation between parents and their newborns is highly recommended during infant hospitalization in the neonatal intensive care unit.

**Objective**

To study the association of a family integrated care (FICare) model compared to standard neonatal care (SNC) on maternal mental health at hospital discharge of their preterm newborn.

**Design**

Prospective observational cohort study (May 2017-January 2020, fAMily Integrated Care in the neonatal ward, the AMICA-study) in mothers of preterm infants.

**Setting**

Multicenter study in level 2 neonatal units in the Netherlands, 1 with the FICare model in single family rooms and 2 control sites with open bay units.

**Participants**

296 mothers (96%) of 309 families included in the AMICA-study provided informed consent.

**Exposure**

FICare model in single family rooms with complete couplet-care for the mother-newborn dyad during maternity and/or neonatal care.

**Main outcome measures**

Maternal mental health, measured using the Parental Stress Scale:NICU (PSS-NICU, primary outcome), Hospital Anxiety and Depression Scale, Post-partum Bonding Questionnaire, Perceived Maternal Parenting Self-efficacy Scale and satisfaction with care (EMPATHIC-N). Parent participation (CO-PARTNER) was assessed as a potential mediator of the association of the FICare model on outcomes with mediation analyses (pre-specified).

**Results**

124/141 (88%) mothers in the FICare model and 115/155 (74%) mothers in SNC responded to questionnaires. Mothers in the FICare model had lower total PSS-

NICU stress scores at discharge (adjusted mean difference -12.24, 95%CI -18.44 to -6.044) than mothers in SNC, specifically less stress from mother-newborn separation (adjusted mean difference -1.273, 95%CI -1.835 to -0.712). Mothers in the FICare model were present more (105/125 (85%) vs 42/115 (37%) for > 8hrs/day, adjusted odds ratio 19.4 95%CI 8.1 to 46.1) and participated more in neonatal care (mean: 46.7 SD: 6.9 vs mean 40.8 SD: 6.7 (max score 62), adjusted mean difference 5.7 95%CI 3.7 to 7.5). Active parent participation was a significant mediator of the association between the FICare model and less maternal depression and anxiety (adjusted indirect effect -0.133, 95%CI -0.226 to -0.055), higher maternal self-efficacy (adjusted indirect effect 1.855, 95%CI 0.693 to 3.348) and better mother-newborn bonding (adjusted indirect effect -0.169, 95%CI -0.292 to -0.068).

**Conclusion and Relevance**

The FICare model in our study was associated with less maternal stress at discharge. The FICare model enables mothers to be more present and participate more in the care for their newborn than in SNC, which is associated with improved maternal mental health outcomes. Future intervention strategies should aim at reducing mother-newborn separation and intensifying active parent participation in neonatal care.

**Trial registration**

https://www.trialregister.nl/trial/6175

# INTRODUCTION

Having a preterm infant (born before 37 weeks of gestation) in the neonatal intensive care unit (NICU) can be a stressful experience, and parents of preterm infants are at a higher risk of developing depression and anxiety postnatally.[195,418,419] Parents can experience infant hospitalization in the NICU as traumatic and can develop post-traumatic stress complaints.[59,420] They are generally assigned to a supportive role during infant hospital stay and often feel insecure or unprepared to care for their infant after discharge.[49,89,380,421] Additionally, due to hospital policies and accommodations, parents often cannot be with their infant continuously leading to parent-infant separation during maternal and neonatal care.[31,68,186,187]

Changing hospital care culture, enabling parents to actively participate in care, being present continuously and achieving closeness can be challenging.[49,192,214,221] Previous studies have shown that participation in care, with a family integrated care (FICare) approach, can alleviate maternal stress at discharge.[55,69] Also, in two systematic reviews and meta-analyses, NICUs with single family rooms (SFRs) were associated with health benefits for infants[147] and parents, specifically stress reduction in mothers[68] possibly through increased parental presence, skin-to-skin care or involvement in care.[134] However, the exact mechanisms on how FICare and SFRs accommodate a reduction in stress, and which exact domains of participation in care are promoted and need to be enforced remains to be elucidated.[68,422] Also, as not all units are able to change their architectural setting to a SFR design and because FICare can be implemented in open bay units, it is important to discern if active parent participation is a mediator for maternal mental health outcomes (such as anxiety and depression).

# OBJECTIVE

To explore the association of a FICare model in single family rooms compared to standard neonatal care (SNC) in open bay units and stress in mothers of preterm infants. Secondary, we determined if the FICare model was associated with improved outcomes in maternal depression, self-efficacy, mother-newborn bonding and satisfaction with care. We studied active participation in neonatal care as a potential mediator in the pathway between the FICare model and maternal mental health outcomes.

# METHODS

This study is part of the fAMily Integrated Care in the neonatal ward study (AMICA-study[236], Appendix, eMethods), a prospective observational cohort study comparing an innovative neonatal care model (FICare model) with standard neonatal care in open bay units. The primary outcome is neurodevelopment in preterm infants at 2 years of corrected age. Also mental health in parents is studied in the short and longer term. The study was registered on the 23rd of December 2016 in the Netherlands Trial Registry NL6175.[236] Hospital architectural design limited randomization between hospitals and randomization within hospitals was impossible, with risks of cross-contamination. Therefore we included infants consecutively that were admitted to participating units. We used the TREND-checklist for non-randomized studies[151] and the AGReMA-SF for report of mediation analyses.[423,424] This study was approved by the medical ethical review committee of Medical Research Ethics Committees United Nieuwegein, the Netherlands.

All infants born in or transferred to the level 2 neonatal units participating in the study (1 exposure and 2 control sites) in the Netherlands were eligible. All participating units had a comparable patient population.

Preterm infants (<37 weeks' gestation) with a hospital stay >7 days and their parents were included after the parents provided written informed consent. For this study we analyzed the mothers of the families. Exclusion criteria were severe psychosocial problems (parents with active psychiatric illness (i.e. psychosis) and/or under supervision of child services), parents non-proficient in Dutch or English, infant congenital abnormalities likely to influence neurodevelopment, or if death of an infant occurred (see Appendix, eMethods).



### Exposure: the FICare model

The exposure setting comprised several aspects: implementation of FICare principles[55], with active parent participation and collaboration between the parents and health care team, and integration of the neonatal and maternity ward enabling couplet-care in SFRs.[68,200] The Mother-Child center was opened in October 2014 in a large teaching hospital in Amsterdam, the Netherlands with 53 SFRs and full integration of maternity and neonatology services.[200] Mothers and infants always stay together in one SFR and never have to be separated as couplet-care can be provided when both need medical care. Fathers can sleep in the SFR and are welcome 24 hours a day.[200] In these rooms prenatal monitoring, labour and postnatal care can be provided for mother and infant together (Figure1a-c).

**Figure 1a. Complete couplet care set up**

A Single-Family Room for highly complex maternity and neonatal Level 2 Care. Women and their newborns will remain in this suite for as long as both require specialized care, or at least for 7 days if the newborn requires specialized care. Fathers, too, can be present continuously. If after 7 days and one of them no longer needs specialized care, the woman and the newborn are transferred to a smaller single-family room, a room for highly complex maternity care and neonatal level 1 care or a room for neonatal level 2 care (Figure 1b). All single-family rooms provide rooming-in facilities for one parent/partner.[200] Copyrights Audiovisuele Zaken, OLVG, Amsterdam, The Netherlands, June 2020.

**Figure 1b. Single family room for neonatal level 2 care**

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**Figure 1c. Family participation in the single family room**

Depicted is a family with twin infants born at a gestational age of 32 weeks, together with a doctor and nurse specialized in neonatal care. The family stays continuously together in a single family room in our integrated neonatal-maternity ward. This enables both parents to participate, as equal partners in the medical team, in the care and medical decision making for their infants during hospital stay. Copyrights Audiovisuele Zaken, OLVG, Amsterdam, The Netherlands, June 2020.

Additionally, a concomitant FICare program was implemented in which parents are trained to be their infant's primary caregiver, while nurses support, teach, coach and counsel parents and perform specific nursing tasks[55,89,328] and necessary specialized medical care, such as cardiorespiratory monitoring, intravenous fluids or antibiotics, placing nasogastric tubes, non-invasive and short-term ventilation and phototherapy. Parents are encouraged but not obliged to actively participate in their infant's care and be present 6-8 hours per day.[55] Parents actively participate as much as they feel comfortable with in neonatal care by (for instance, and not limited to) providing feedings by nasogastric tube, bottle or breast, providing skin-to-skin care, weighing and temperature regulation. Family-centered rounds were implemented including parents on medical rounds, involving them in patient management, and enabling them to hear first-hand the developments in their infant's condition. Parents could provide information on their infant's general wellbeing, ask questions and participate in shared decision making.[154,425]

## Control group (standard neonatal care)

SNC in open bay units (OBUs) was provided in two different level 2 neonatal units in Alkmaar and Amsterdam, The Netherlands. These units have an open configuration with newborns staying together in one unit (with a maximum of approximately 18 infants admitted simultaneously, see Figure1d). These OBUs are close to the maternity ward, but physically separated. Infants who require high-intensive care, tubefeeding, cardiorespiratory monitoring, respiratory support, antibiotics or phototherapy are admitted to these wards. Adjacent to these wards, maternity wards are present, where mothers can stay up to 7 days after giving birth. Parents can be with their infant, provide skin-to-skin care, (breast)feeding and participate in their infant's care. Medical rounds are done in a separate room without parents. Nurses provide routine care. The OBUs cannot provide the necessary facilities for 24h presence, especially a place to sleep or rest for the mother. Facilities in the OBU include: comfortable chair at the bedside, possibility to express breastmilk with a mechanical device near the infant and separate rooms to have conversations with the medical team.

**Figure 1d. Open bay unit with standard neonatal care**



## Outcomes and mediators

The predefined primary outcome for this study in mothers was stress as measured by the Parental Stress Scale: NICU (PSS-NICU) questionnaire[241] at discharge. Parents rate their experiences on stressors associated with the hospitalization of their child on a

5-point rating scale ranging from “not at all stressful” (0) to “extremely stressful” (5)[241] (maximum score 130, with higher scores indicating more stress). Secondary maternal mental health outcomes included: measurements at discharge of maternal depressive symptoms and anxiety (Hospital Anxiety and Depression Scale, maximum score 42, with higher scores indicating more depressive symptoms)[242], parent self-efficacy (Perceived Maternal Parenting Self-efficacy Scale; maximum score, 80, with higher scores indicating more self-efficacy)[243], impaired mother-newborn bonding (Post-partum Bonding Questionnaire; maximum score, 125, with higher scores indicating more impaired mother-newborn bonding)[245], satisfaction with care and empowerment (EMpowerment of PArents in THe Intensive Care–Neonatology; maximum score, 6, with higher scores indicating more satisfaction).[244] Mothers filled out how they participated and collaborated with health care staff in neonatal care using the CO-PARTNER tool (maximum score, 62, with higher scores indicating more participation and collaboration in neonatal care[422]).

Also, mothers filled out a general questionnaire with details on their education, current job and the cultural background they identified most with (classified by the participant). To improve response rates, mothers were reminded up to 2 times (7 and 14 days after initial questionnaires were sent). For an elaborate explanation see Appendix, eMethods and eTable 1.

### Statistical analyses

Two samples t-tests were used to compare continuous variables between the FICare group and SNC group. For non-normally distributed variables Mann-Whitney U tests were used. To analyze proportions between groups the $\chi^2$-test was used. If expected cell frequency was <5, Fisher’s exact test was used.



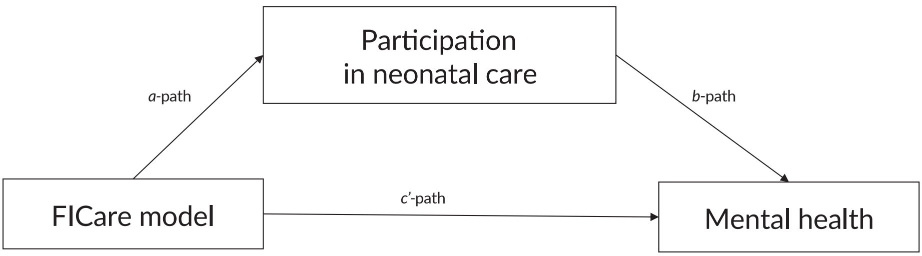
Baseline characteristics between mothers with and without outcomes variables at discharge were compared. We assumed that the data were missing-at-random. The proposed guidance as explained by Sterne et al. was applied for missing data[158] and we applied the multivariate imputation by chained equations (mice) procedure with parcel mean summary scores to missing data at the item level.[249] All variables used in the analyses were included in the imputation model, as well as auxiliary variables related to the probability of missing data or to the variables with missing data itself. Variables that were multicollinear with other included variables were excluded from the imputation model. For all data sets, we performed 10 imputations and 50 iterations to obtain imputed data sets. Convergence was checked graphically with convergence plots. All analyses were performed on the imputed datasets and results were pooled by using Rubin’s Rules.[160]

We performed multivariable linear and logistic regression in imputed datasets estimating crude and adjusted associations between the FICare model and maternal mental health outcomes. Logarithmic transformations were applied to normalize skewed distributions, or if unsuccessful, dichotomization. Potential confounders and effect modifiers were identified from the literature and assessed using statistical analyses (see Appendix, eMethods).

We hypothesized that the FICare model (exposure) transmits its association on maternal mental health outcomes (the outcome) at discharge (partially) through active parent participation (the mediator, CO-PARTNER score, Figure 2). Mediation analyses on the imputed dataset were therefore applied to analyze, identify and explain the underlying mechanisms of the observed total effect of the FICare model on mental health outcomes in mothers (i.e. the *c*-path)[161] also in the absence of a significant total effect (*c* path) as described before.[426]

In addition to the total association model, two linear regression models were fitted. In single mediator models total parent participation was included as an individual potential mediator of different mental health outcomes in mothers (Figure 2). In the first regression model the effect of the FICare model on the mediator was estimated (*a*-path). In the second regression model the effect of the mediator (participation) on outcomes (*b*-path) and the direct effect of the FICare model on outcomes (*c'*-path) were estimated. We calculated the indirect effect (the amount of mediation) in the single mediator models as the product of the *a*- and *b*-coefficients. Crude and adjusted mediation analyses were performed. In the adjusted analyses, confounders were added to all models. We used bootstrap 95% percentile confidence intervals based on 1,000 bootstrap resamples around the indirect effects.[162,262]

**Figure 2. Parent participation as a mediator of the effect of the FICare model on mental health outcomes in mothers**



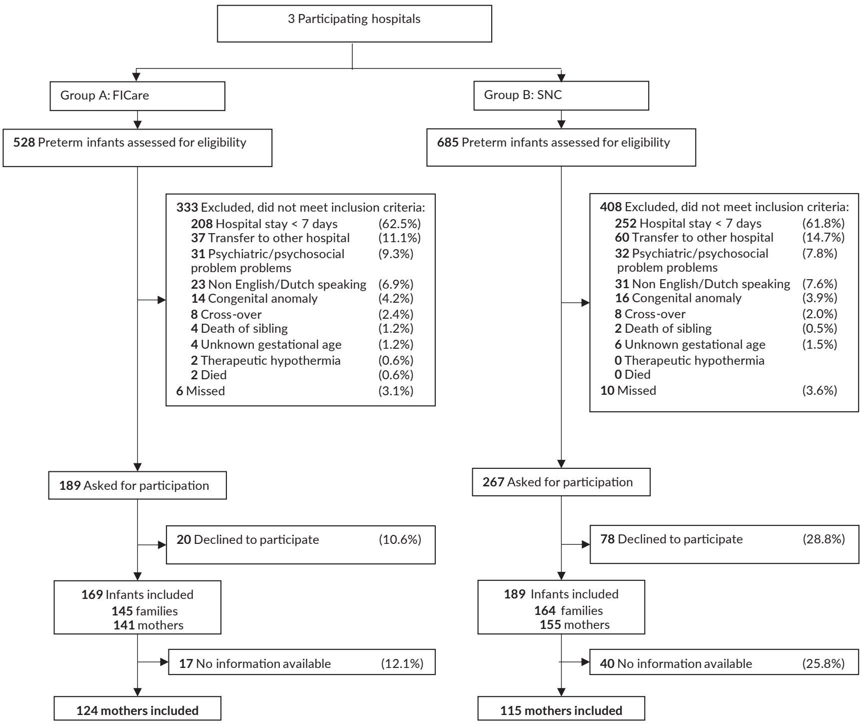

FICare: family integrated care

We used R for statistical analyses (version 3.6.1)[163] and specifically for multiple imputation the 'mice'-package[159], for analyzing missing data patterns the 'VIM'-package[164], and for the 95% percentile bootstrap confidence interval the 'boot'-package.[167] For all tests, a *p*-value of less than 0.05 was considered statistically significant. Data analysis was performed from January to April 2021.

## RESULTS

From the 19th of May 2017 through the 8th of January 2020, we recruited 309 families (145 in FICare and 164 in SNC), encompassing 358 infants and their parents (Figure 3). During the recruitment period, one of the control sites changed to a double-bed occupancy with SFR-like design and FICare practices, and discontinued recruitment of control patients in March 2019. 296 (96%) mothers consented to participate in the study regarding their mental health (141 in FICare and 155 in SNC). 239/296 mothers (81%), filled out surveys and were analyzed (124/141 (88%) in the FICare model (mean age 33.3 (4.0) years) and 115/155 (74%) in SNC (33.3 (4.1) years, see Appendix eTable 2-5 for response rates and missing data).

**Figure 3. Flowdiagram of study**



Baseline characteristics for mothers were similar between the exposure and control group, except for infant gestational age which was lower in the FICare model (median 32 weeks 1 days (IQR: 29+3 to 34+5) vs 34 weeks (32+2 to 34+6), p<0.001, Mann-Whitney U Test) (Table 1). Infants were also less often born in the level 2 facility in the FICare model compared to SNC (53/124 (43%) vs 80/115 (70%) p<0.001, Chi-square test).

Overall, mothers in the FICare model had significantly lower total NICU stress scores (adjusted mean difference -12.24, 95%CI -18.44 to -6.044), lower stress from infant behavior, sights and sounds (adjusted mean difference -5.819, 95%CI -10.29 to -1.350) and lower stress scores due to parental role alteration (adjusted mean difference –6.423, 95%CI -8.910 to -3.937) at discharge compared to mothers in SNC (Table 2).

In the PSS-NICU questionnaire, 34/188 mothers (18.1%) scored the item "stress due to separation from their infant" as "extremely stressful" (Appendix, eTable6), mostly by mothers in SNC (24/34; 70.6%). The mean stress score on this item was significantly lower in the FICare model (2.1 (SD 2.0)) compared to mothers with infants admitted to SNC (3.3 (SD 1.6)) also after adjusting for confounders (adjusted mean difference -1.273, 95%CI -1.835 to -0.712).

## Participation during hospital stay

Mothers in the FICare model were present more than mothers in SNC (median 20 (IQR: 9-24)) versus 6 (IQR: 4-12)) hours/day). 105/124 mothers (85%) were able to be present for at least 8 hours in the FICare model compared to 42/115 (37%) in SNC (Table 2, adjusted odds ratio 19.35 95%CI 8.130 to 46.08). Mothers in the FICare model participated more in care of their infant (adjusted mean difference 5.618, 95%CI 3.705 to 7.532) compared to SNC (Table 2). Participation was higher in mothers in the FICare model compared to SNC, specifically within daily care, medical care (including tubefeeding, monitoring of the infant, regulation of visitation to the infant and participating in daily rounds), advocacy and leadership, time spent with the infant, and comforting of the infant. In the FICare model, mothers required less information compared to mothers in SNC.

## Mediation analyses of active parent participation on maternal mental health outcomes

With mediation analyses we estimated the indirect effect (the *ab* path) of the FICare model on maternal mental health outcomes through active parent participation. We also estimated the direct effect of the FICare model on maternal mental health outcomes which was **not** explained by increased active parent participation (through the *c'* path).

Increased active maternal participation was a significant mediator of the association between the FICare model and less maternal depression and anxiety (adjusted indirect effect -0.133, 95%CI -0.226 to -0.055, *ab* path), better mother-newborn bonding (adjusted indirect effect -0.169, 95%CI -0.291 to -0.068, *ab* path) and higher maternal self-efficacy (adjusted indirect effect 1.855, 95%CI 0.693 to 3.348, *ab* path) at discharge (Table 3). In other words, the higher active maternal participation in the FICare model (adjusted *a* path: 5.618, SE 0.969) was associated with lower depressive symptomatology (adjusted *b* path: -0.024 SE 0.007), better mother-newborn bonding scores (adjusted *b* path: -0.030 SE: 0.009), and higher self-efficacy scores (adjusted *b* path: 0.330 SE 0.091). No beneficial direct effects (*c'* paths) were found of the FICare model on maternal depression and anxiety, mother-newborn bonding and maternal self-efficacy.

The FICare model was associated with less stress for mothers at discharge compared with mothers in SNC. Increased active parent participation in the FICare model was a potential mediator of this association, but did not reach statistical significance (adjusted indirect effect -2.148, 95%CI -5.045 to 0.201, *ab* path, Table 3). The direct effect (*c'* path) of the FICare model on maternal NICU stress remained large after adjustment for active parent participation (adjusted *c'* path -10.09 SE: 3.397).

Parent satisfaction was not different between the FICare model and SNC, and increased active parent participation was not a mediator of the association between the FICare model and satisfaction with care (adjusted indirect effect 0.036, 95%CI -0.012 to 0.095).

## DISCUSSION



This study shows that mothers of preterm infants experience less stress at discharge when admitted to a setting with FICare in SFRs compared to SNC. Mothers in the FICare model were able to be present more and participate more in neonatal care, which was associated with improved mental health outcomes including less depression, better mother-newborn bonding and higher self-efficacy.

In concordance with previous research, our results indicate an association between mother-newborn separation and high stress levels in mothers of preterm infants[31,49] admitted to SNC settings. Mother-newborn separation is one of the main challenges health care professionals currently encounter when caring for mothers and infants postnatally, especially when both need medical care. Also during the COVID-pandemic it is again becoming apparent that restrictive policies and mother-newborn separation are of great concern.[427] Parents report that this limits their ability to bond with their infant, to participate in care, negatively impacting breastfeeding as well.[219,428,429]

For NICU stress, a large direct association (*c'* path) of the FICare model - independent of active maternal participation- was present. This could indicate that the architectural design with complete couplet-care for the mother-newborn dyad in single family rooms was an important factor associated with less maternal stress at discharge, as has been shown before.[68] The architectural design may have been less important for the other maternal mental health outcomes, since we found no direct beneficial association (*c'* path) for these outcomes. However, increased active maternal participation was a significant mediator of the association between the FICare model and less maternal depression and anxiety, better mother-newborn bonding and higher maternal self-efficacy. These findings suggest that for maternal depression, mother-newborn bonding and maternal self-efficacy specific attention should be pointed towards active maternal partnership and collaboration in neonatal care. Improving active maternal participation and collaboration in neonatal care is feasible independent of the architectural design, as the FICare-methodology was initially developed in an OBU.[56,93,430]

Strengths of this study are that we used a validated questionnaire (CO-PARTNER)[422] to evaluate maternal participation in neonatal care, which has not been done as rigorously before.[134,422] We used advanced statistical techniques for missing data and mediation analyses. We included families with infants within a range of all gestational ages, reflecting reality of a level 2 neonatal unit, and high response rates were achieved.

Future research should focus on both parental and neonatal outcomes after discharge as effects of NICU hospitalization on infants (i,e, neurodevelopment[147]) and parents (i.e. traumatic stress[431]) could persist. Also more studies should explore the influence of hospitalization of a preterm infant on fathers, as they too can experience adverse outcomes.[61,432]

Additionally, research should focus on an exact definition of “zero separation” in this context, as one can still feel emotionally connected without being physically present. Research studies could for instance qualitatively focus on the perception of emotional closeness and the pathways towards emotional closeness that might be facilitated in our FICare model from parents’ perspectives.[205]

**Limitations**

As this was a non-randomized study, several limitations were inherently present. We had different enrollment numbers between the FICare model and SNC settings. This was mainly due to non-consent in SNC, and not due to missed participants (these numbers were similar between settings). Also, potential baseline differences were present, specifically for gestational age. However, despite this, mothers in the FICare model still experienced less stress due to parental role alteration and specifically less stress from being separated from their infant.

Additionally, the potential causality which might be suggested with mediation analysis, should also be considered. Mothers who are less depressed/better bonded/highly self-efficient might also participate more in care, and health care professionals should consider this when implementing programs aimed at increasing parent participation.

## Conclusion

These findings suggest that setting up level 2 neonatal units with a FICare model in single family rooms with complete couplet-care for the mother-newborn dyad is associated with reduced maternal stress at discharge compared to SNC in OBUs with separate maternity care. In this FICare model, mothers can participate and collaborate more in neonatal care, which is associated with ameliorated maternal mental health. For future ward reconfigurations zero-separation between mothers and their newborn should be strived for. However, independent of the architectural design of the neonatal unit, mothers should be allocated as active partners in neonatal care.

**Table 1. Baseline characteristics mothers**

| Characteristic | FICare group (n=124) | SNC group (n=115) | P-value |
|---|---|---|---|
| Age (years, mean (SD)) | 33.3 (4.0) | 33.3 (4.1) | 0.56 |
| University degree (No (%)) | 108/113 (95.6%) | 89/100 (89%) | 0.19 |
| Paid job (No (%)) | 91/113 (80.5%) | 85/100 (85%) | 0.73 |
| Identifies with Dutch cultural background (No (%)) | 87/115 (76%) | 89/102 (87%) | 0.05 |
| Stress of pregnancy (mean (SD), max score 5) | 2.3 (1.3) | 2.3 (1.2) | 0.95 |
| Stress of birth (mean (SD), max score 5) | 3.0 (1.4) | 2.8 (1.3) | 0.24 |
| Pre-eclampsia (No (%)) | 29/120 (24%) | 21/114 (18%) | 0.36 |
| HELLP syndrome (No (%)) | 5/124 (4%) | 7/112 (6%) | 0.70 |
| Use of psychofarmaca (No (%)) | 2/124 (2%) | 3/115 (3%) | >0.99[a] |
| Gestational age (wks, median (IQR), range) | $32^{+1}$<br>($29^{+3}$ – $34^{+5}$)<br>($24^{+1}$ – $36^{+6}$) | $34^{+0}$<br>($32^{+2}$ – $34^{+6}$)<br>($24^{+1}$ – $36^{+6}$) | <0.001 |
| Born < 32 weeks of gestation (No (%)) | 60/124 (48%) | 25/115 (22%) | <0.001 |
| Inborn infant (born in level 2 hospital) (No (%)) | 53/124 (43%) | 80/115 (70%) | <0.001 |
| Singleton (No (%)) | 103/124 (83%) | 102/115 (89%) | 0.29 |
| Vaginal delivery (No (%)) | 64/124 (52%) | 60/115 (52%) | 0.30 |
| First child upbringing? (No (%)) | 81/113 (72%) | 65/95 (68%) | 0.13 |
| Plan for upbringing *Together with partner* (No (%)) | 107/113 (95%) | 86/94 (91%) | 0.11 |
| Total stress at admission (mean (SD), *max score 130*) | 55.7 (22.7) | 56.1 (21.8) | 0.88 |
| Depression and anxiety score at admission (median (IQR), *max score 42*) | 10 (8 – 14) | 12 (7 – 24) | 0.46 |

[a]: Fisher exact test Data are n/N (%), mean (SD) or median (IQR). Denominators differ because of missing data (see Appendix, eTable 5). Range (min – max), FICare: family integrated care, HELLP: hemolysis, elevated liver enzymes, and a low platelets (complication of pregnancy), IQR: interquartile range, max: maximum, No: number, SD: standard deviation, SNC: standard neonatal care, wks: weeks

**Table 2. Maternal participation in neonatal care during hospital stay and mental health outcomes at discharge**

| | FICare (n = 124) | SNC (n = 115) | Mean difference (95%CI) | p-value | Adjusted mean difference (95%CI)[a] | p-value |
|---|---|---|---|---|---|---|
| ***During hospital stay*** | | | | | | |
| **Presence (hours per day, median (IQR))** | 20 (9 – 24) | 6 (4 – 12) | -[b] | - | -[b] | - |
| **Presence (>8hrs per day, n (%))** | 105 (84.7) | 42 (36.5) | 9.578 (4.988; 19.39)[c] | <0.001 | 19.35 (8.130; 46.08)[c] | <0.001 |
| ***Total participation (mean (SD), max score 62)*** | 46.7 (6.9) | 40.8 (6.7) | 5.917 (4.126; 7.708) | <0.001 | 5.618 (3.705; 7.532) | <0.001 |
| **Domain 1** *Participation in daily care (mean (SD), max score 22)* | 16.5 (4.0) | 15.4 (3.1) | 1.043 (0.081; 2.006) | 0.03 | 0.953 (-0.061; 1.969) | 0.07 |
| **Domain 2** *Participation in medical care (mean (SD), max score 8)* | 4.7 (1.8) | 3.5 (1.5) | 1.196 (0.754; 1.638) | <0.001 | 1.037 (0.582; 1.492) | <0.001 |
| **Domain 3** *Information gathering (mean (SD), max score 3)* | 2.3 (0.8) | 2.5 (0.6) | -0.190 (-0.402; 0.022) | 0.08 | -0.311 (-0.537; -0.085) | 0.008 |
| **Domain 4** *Advocacy and leadership (mean (SD), max score 3)* | 2.2 (1.0) | 1.5 (1.1) | 0.692 (0.417; 0.965) | <0.001 | 0.636 (0.357; 0.916) | <0.001 |
| **Domain 5** *Time spent with infant (mean (SD), max score 12)* | 8.3 (2.4) | 6.1 (2.8) | 2.157 (1.412; 2.902) | <0.001 | 2.297 (1.529; 3.065) | <0.001 |
| **Domain 6** *Comforting the infant (mean (SD), max score 14)* | 12.7 (1.3) | 11.7 (1.7) | 1.021 (0.514; 1.528) | <0.001 | 1.010 (0.502; 1.519) | <0.001 |
| ***Outcomes at discharge*** | | | | | | |
| NICU stress (mean (SD), *max score 130*) | 47.2 (22.2) | 57.0 (22.2) | -9.737 (-16.01; -3.465) | 0.003 | -12.24 (-18.44; -6.044) | <0.001 |
| *Behaviour and sights and sounds* (mean (SD), *max score 95*) | 34.5 (16.2) | 38.6 (15.2) | -4.022 (-8.721; 0.677) | 0.09 | -5.819 (-10.29; -1.350) | 0.01 |
| *Parental role alteration* (mean (SD), *max score 35*) | 12.7 (8.1) | 18.4 (9.1) | -5.715 (-8.239; -3.191) | <0.001 | -6.423 (-8.910; -3.937) | <0.001 |
| *Being separated from my baby* (mean (SD), *max score 5*) | 2.1 (2.0) | 3.3 (1.6) | -1.174 (-1.698; -0.651) | <0.001 | -1.273 (-1/835; -0.712) | <0.001 |

**Table 2. Maternal participation in neonatal care during hospital stay and mental health outcomes at discharge (continued)**

| | FICare (n = 124) | SNC (n = 115) | Mean difference (95%CI) | p-value | Adjusted mean difference (95%CI)[a] | p-value |
|---|---|---|---|---|---|---|
| Anxiety and depression (median (IQR), *max score 42*) | 9.8 (5.3 – 15.3) | 10.1 (4.8 – 15.5) | -0.062 (-0.252; 0.128)[d] | 0.52 | -0.117 (-0.308; 0.075)[d] | 0.23 |
| Self-efficacy (mean (SD), *max score 80*) | 63.7 (8.9) | 62.7 (9.0) | 1.002 (-1.357; 3.361) | 0.40 | 0.916 (-1.532; 3.364) | 0.46 |
| Impaired mother-newborn bonding (median (IQR), *max score 125*) | 10.2 (4.1 – 16.3) | 9.3 (4.3 – 14.4) | 0.142 (-0.076; 0.361)[d] | 0.20 | 0.097 (-0.130; 0.324)[d] | 0.40 |
| Satisfaction with care (median (IQR), *max score 6*) | 5.6 (5.3 – 5.9) | 5.6 (5.2 – 5.9) | 0.018 (-0.104; 0.140)[d] | 0.77 | 0.023 (-0.099; 0.146)[d] | 0.71 |

All outcomes are pooled estimates from multiple imputed datasets, Outcomes are calculated from the imputed datasets. [a]adjusted for: Gestational age, gemelli status, education, age, Dutch background, singleton status, stress at birth and first child upbringing, [b] regression estimates could not be calculated due to non-normality also after logarithmic transformation, [c]Odds ratio, [d]after logarithmic transformation,

**Table 3. Mediation analysis of mothers' participation during infant hospital stay and mental health at discharge**

| Outcome | Effect of FICare on mediator (participation) (*a* pathway (SE)) | Effect of mediator (participation) on outcome (*b* pathway (SE)) | Indirect effect (*ab*) | 95% CI of indirect effect (*ab*) | Effect of FICare on outcome (*c'*- pathway (SE)) | Effect of FICare on outcome (*c*-pathway (SE)) |
|---|---|---|---|---|---|---|
| *Crude analyses* | | | | | | |
| Stress | 5.917 (0.908) | -0.393 (0.225) | -2.324 | -5.156 to 0.186 | -7.410 (3.485) | -9.737 (3.167) |
| Self-efficacy | 5.917 (0.908) | 0.343 (0.092) | 2.031 | 0.805 to 3.479 | -1.030 (1.299) | 1.002 (1.196) |
| Satisfaction with care | 5.917 (0.908) | 0.004 (0.004) | 0.024 | -0.025 to 0.078 | -0.006 (0.067) | 0.018 (0.062) |
| Depression and anxiety[a] | 5.917 (0.908) | -0.024 (0.008) | -0.143 | -0.243 to -0.057 | 0.081 (0.105) | -0.062 (0.096) |
| Impaired mother-newborn bonding[a] | 5.917 (0.908) | -0.031 (0.009) | -0.186 | -0.316 to -0.077 | 0.328 (0.120) | 0.142 (0.111) |
| *Adjusted analyses[b]* | | | | | | |
| Stress | 5.618 (0.969) | -0.382 (0.214) | -2.148 | -5.045 to 0.201 | -10.09 (3.397) | -12.24 (3.13) |
| Self-efficacy | 5.618 (0.969) | 0.330 (0.091) | 1.855 | 0.693 to 3.348 | -0.939 (1.322) | 0.916 (1.242) |
| Satisfaction with care | 5.618 (0.969) | 0.007 (0.004) | 0.036 | -0.012 to 0.095 | -0.013 (0.067) | 0.023 (0.062) |
| Depression and anxiety[a] | 5.618 (0.969) | -0.024 (0.007) | -0.133 | -0.226 to -0.055 | 0.017 (0.101) | -0.117 (0.097) |
| Impaired mother-newborn bonding[a] | 5.618 (0.969) | -0.030 (0.009) | -0.169 | -0.292 to -0.068 | 0.267 (0.121) | 0.097 (0.114) |

All outcomes are pooled estimates from multiple imputed datasets, [a]after logarithmic transformation, [b]adjusted for: Gestational age, gemelli status, education, age, Dutch background, singleton status, stress at birth and first child upbringing.

Chapter 10

# Association of a Family Integrated Care Model with Paternal Mental Health Outcomes During Neonatal Hospitalization

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# ABSTRACT

### Importance
During newborn hospitalization in the neonatal unit, fathers often feel anxious and excluded from their child's caregiving and decision-making. Few studies and interventions have focused on fathers' mental health and their participation in neonatal care.

### Objective
To study the association of a family integrated care (FICare) model (in single family rooms with complete couplet-care for the mother-newborn dyad) versus standard neonatal care (SNC) in open bay units with separate maternity care with mental health outcomes in fathers at hospital discharge of their preterm newborn and to study whether parent participation was a mediator of the FICare model on outcomes.

### Design, Setting and Participants
This prospective, multicenter cohort study was conducted from May 2017 to January 2020 as part of the fAMily Integrated Care in the Neonatal Ward Study, at leve-02 neonatal units in the Netherlands (1 using the FICare model and 2 control sites using SNC). Participants included fathers of preterm newborns admitted to participating units. Data analysis was performed from January to April 2021.

### Exposure
FICare model in single family rooms with complete couplet-care for the mother-newborn dyad during maternity and/or neonatal care.

### Main outcomes and measures
Paternal mental health was measured using the Parental Stress Scale:NICU, Hospital Anxiety and Depression Scale, Post-partum Bonding Questionnaire, Perceived (Maternal) Parenting Self-efficacy Scale, and satisfaction with care (Empowerment of PArents in THe Intensive Care -Neonatology). Parent participation(CO-PARTNER-tool) was assessed as a potential mediator of the association of the FICare model with outcomes with mediation analyses (pre-specified).

### Results
Of 309 families included in the fAMily Integrated Care in the Neonatal Ward Study, 263 fathers (85%) agreed to participate; 126 fathers were enrolled in FICare and 137 were enrolled in SNC. In FICare, 89 fathers (71%, mean (SD) age 35.1 (4.8) years) responded to questionnaires and were analyzed. In SNC, 93 (68%; mean (SD) age

36.4 (5.5) years) responded to questionnaires and were analyzed. Fathers in FICare experienced less stress (adjusted beta -10.02,95%CI -15.91 to -4.130,p=0.001) and had higher participation scores (adjusted beta 3.424,95% CI 0.860 to 5.988,p=0.009) compared with those in SNC. Participation mediated the beneficial association of the FICare model with fathers' depressive symptoms (-0.051,95%CI -0.133 to -0.003), and bonding with their newborns (-0.082,95%CI-0.177 to -0.015)

**Conclusion and Relevance**

These findings suggest that the FICare model is associated with decreased paternal stress at discharge and enables fathers to be present and participate more than in SNC, thus improving paternal mental health. Supporting fathers to actively participate in all aspects of newborn care should be encouraged regardless of architectural design of the neonatal unit.

**Trial registration**

https://www.trialregister.nl/trial/6175

# INTRODUCTION

Parents can experience hospitalization of their preterm newborn in the neonatal intensive care unit (NICU) as very stressful.[49,433] Integrating the family as a relevant and irreplaceable part of the health care team and creating an environment welcoming continuous parental presence[68] and active participation in neonatal care, or family integrated care (FICare) has been shown to be beneficial for mothers and their newborns.[55,69,155]

In addition to the mothers, fathers (or partners) also, play an important role during newborn hospital stay and newborn development.[434] In animal models, paternal presence early in life is associated with increased survival[435] and improved social behaviors and emotional functions in offspring later in life.[436] During the NICU stay of their newborn, human fathers often feel excluded from their newborn's caregiving and decision-making.[433] They are expected to support mothers and participate in care of their newborn, but they can also experience trauma, anxiety and depression following preterm birth.[437–439] They can struggle to combine a sustained presence in the NICU while maintaining employment and domestic responsibilities outside the NICU.[440] Additionally, fathers can develop feelings of insecurity, helplessness, and a lack of control if they are not involved in their newborns' care.[289] Among mothers, FICare is associated with less stress[69], but it is unknown through which mechanisms. For fathers, little research has been conducted concerning their perinatal experiences in the event of prematurity and, specifically, studying the association of the neonatal care setting and father's participation in newborn care with paternal mental health outcomes.

The primary objective was to study the association of the FICare model in single family rooms with complete couplet-care for the mother-newborn dyad versus standard neonatal care (SNC) in open bay units with mental health outcomes (stress, anxiety, depression, impaired father-newborn bonding, self-efficacy and satisfaction) among fathers at discharge of their preterm newborn. The secondary objective was to study whether parent participation was a mediator of the association of the FICare model on paternal mental health.

# METHODS

## Study design

This study is part of the fAMily Integrated CAre in the neonatal ward study (eAppendix 1), a prospective, observational, cohort study comparing the FICare model with SNC in open bay units. The primary outcome is neurodevelopment in preterm newborns at the corrected age of 2 years.[236] Mental health outcomes in parents are also studied in the short and long term. This study follows the Transparent Reporting of Evaluations with Nonrandomized Designs (TREND)-checklist and A Guideline for Reporting Medioation Analyses of Randomized Trials and Observational Studies (AGReMA-SF).[423,424] This study was approved by the medical ethical review committee of Medical Research Etrhics Committees United Nieuwegein, the Netherlands.

All newborns born in or transferred to level 2 neonatal wards participating in the study (1 exposure and 2 control sites) in the Netherlands were eligible. Preterm newborns (<37 weeks' gestation) with a hospital stay longer than 7 days and their parents were included after the parents provided written informed consent. For this study, we analyzed the fathers of the families. We also included same-sex couples as we recognize and respect that there are people having children who may not identify as father or mother. For the sake of clarity, we use the term *fathers* for partners of the newborn's mother who will assume a parental role. Exclusion criteria were severe psychosocial problems (parents with active psychiatric illness (i.e. psychosis) and/or under supervision of child services), parents nonproficient in Dutch or English, newborn congenital abnormalities likely to influence neurodevelopment, or if death of a newborn occurred (see eAppendix 1). Figure 1 Shows the study enrollment flow chart.

## Exposure (FICare model)

Within a large teaching hospital with a level-2 neonatal unit in Amsterdam, the Netherlands, an innovative FICare model was set up including complete mother-newborn couplet-care in single family rooms with rooming-in facilities with a concomitant participation program for parents and recurring education for staff. In this setting, integration between maternal and neonatal services was achieved for all newborns and their families[200]; mothers never had to be separated from their newborns during hospital stay when neonatal and/or maternity care was needed (couplet-care). Fathers could be continuously present with the family during obstetric, maternity and neonatal care (eFigure 1-3 in the Appendix). In addition, parents were trained to be the primary caregiver of their newborn, and nurses supported, instructed and counseled parents.[89] Parents were invited but not obligated to be present more than 8 hours per day, and rooming-in facilities were present.[262]

Parents were actively encouraged to participate in all aspects of their newborn's care as much as they felt comfortable with, such as (but not limited to) providing feedings by nasogastric tube, breast or bottle; providing skin-to-skin care; weighing; and regulating temperature control. Family-centered rounds were implemented and included active parental participation in shared decision-making on daily medical rounds and involvement in the process of patient management.[55,154] In addition, parents received group education sessions to learn on all aspects concerning (preterm) newborn and family health.[55,328]

## Control group (SNC)

Two different teaching hospitals with level-2 neonatal units in Amsterdam and Alkmaar, the Netherlands, were control centers in the study. Within these centers maternity and neonatal care services were separated from each other. Ill or preterm newborns born at less than 35 weeks of gestation, weighing less than 2000 grams or in unstable condition were transferred to the neonatal unit. Maternity care was delivered in a ward separate from the neonatal ward. The neonatal units were set-up with open bay units (eFigure 4 in the Appendix). Each incubator was separated by a curtain and had a chair available for parents. Nurses involved parents as much as possible in the care of their newborn. Parents could sign up for weekly updates with the pediatrician. Daily rounds were performed between the nursing staff and pediatrician, without the presence of the family. Nurses usually updated parents after decisions were made during daily rounds. No facilities were present for parents to room-in with their newborn during hospital stay.

## Data collection

Included fathers were asked to fill out mental health-related questionnaires at admission and discharge regarding stress (Parental Stress Scale: NICU (PSS-NICU); maximum score, 130, with higher scores indicating more stress)[241], anxiety and depression (Hospital Anxiety and Depression Scale (HADS); maximum socre, 42, with higher scores indicating more depressive symptoms))[242], parent self-efficacy (Perceived (Maternal) Parenting Self-efficacy Scale (PMP-SE); maximum score, 80, with higher scores indicating more self-efficacy)[243], and impaired parent-newborn bonding (Postpartum Bonding Questionnaire (PBQ); maximum score, 125, with higher scores indicating more impaired parent-newborn bonding).[245] Fathers also completed questionnaires regarding satisfacdtion with care at hospital discharge (Empowerment of Parents in The Intensive Care-Neonatology (EMPATHIC-N), maximum score, 6, with higher scores indicating more satisfaction)[244] and how they participated and collaborated with health care staff in neonatal care using the CO-PARTNER[422] tool (maximum score, 62, with higher scores indicating more participation and collaboration in neonatal care, see Appendix

1, eTable 1 and eTable 2 for an elaboration and sample size calculations). Finally, fathers completed a general questionnaire with details on their education, current job and the cultural background they identified most with (classified by the participant), smoking, alcohol, and recreational drug use. To improve response rates, fathers were reminded up to 2 times (7 and 14 days after initial questionnaires were sent).

## Statistical analyses

We performed independent *t* tests for normally distributed, or Mann-Whitney *U* tests for non-normally distributed data. Chi-square ($\chi^2$) tests were used to test for differences in binary outcomes. If expected cell-counts were 5 or lower, we calculated differences with the Fisher's exact test. All tests were 2-sided.

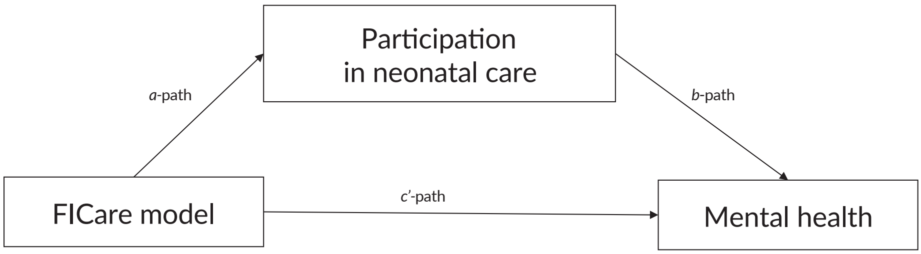
Baseline characteristics between fathers with and without outcome variables at discharge were compared. We assumed that the data were missing-at-random (MAR). We used the proposed guidance as explained by Sterne[158] for missing data and applied the multivariate imputation by chained equations (mice) procedure with parcel summary scores to missing data at the item level.[249] All variables used in the analyses were included in the imputation model, as well as auxiliary variables related to the probability of missing data or to the variables with missing data itself. Variables that were (multi) collinear with other included variables were excluded from the imputation model. For all datasets, we performed 20 imputations and 50 iterations to obtain imputed datasets (see eAppendix 1). Convergence was checked graphically with convergence plots. All analyses were performed on the imputed datasets and results were pooled by using Rubins Rules.[250]

To study associations between the FICare model and outcomes in fathers, we performed multivariable linear or logistic regression in imputed datasets. For nonnormally distributed outcome data, we first applied a (natural) logarithmic- or square root transformation to obtain normal distribution, or if unsuccessful, dichotomized outcomes.

Potential confounders and effect modifiers were identified from the literature and assessed using statistical analyses (see eAppendix 1).

To study parent participation as a potential mediator of the observed association of the FICare model with mental health (i.e. the *c*-path)[161] we performed mediation analyses on the imputed dataset.[161,262] In addition to the total association model, 2 linear regression models were fitted. In single mediator models, total parent participation was included as individual potential mediator of different mental health outcomes in fathers (Figure 1). In the first regression model the association of the FICare model with the mediator was estimated (*a*-path). In the second regression model the association of the mediator with outcomes (*b*-path) and the direct effect size of the FICare model with outcomes (*c'*-path) were calculated. Crude and adjusted mediation analyses were performed. In the adjusted analyses, confounders were added to all models. We calculated the indirect effect size as the product of the *a* and *b* coefficients. We estimated bootstrap 95% percentile confidence intervals based on 1000 bootstrap resamples around the indirect effects sizes.[162,262]

**Figure 1. Parent participation as a mediator of the effect of the FICare model on mental health outcomes in fathers**

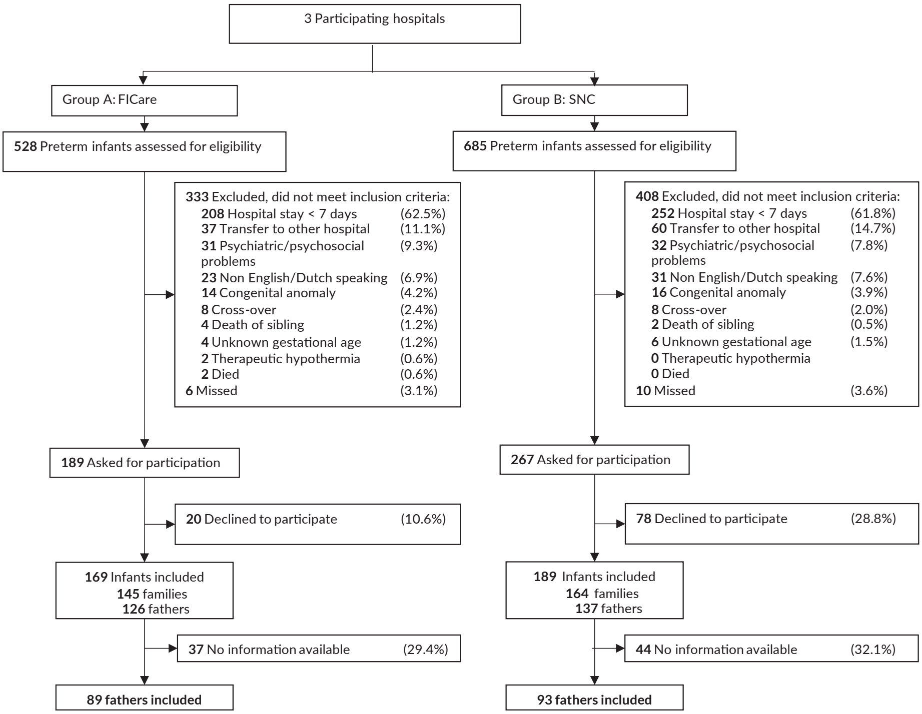



FICare: family integrated care

We used R statistical software version 3.6.1(R Project for Statistical Computing)[163] including the 'mice'-package[248] for multiple imputation, and the 'boot'-package[167] for the 95% bootstrap confidence intervals. For all tests, $P< 0.05$ was considered statistically significant. Data analysis was performed from January to April 2021.

# RESULTS

**Figure 2. Flowdiagram of study**




FICare: family integrated care, SNC: standard neonatal care

A total of 309 families were included in this study, with 358 newborns and 559 parents (296 mothers and 263 fathers, see Figure 2). 126 fathers consented to participate in the FICare model and 137 fathers participated in SNC. 89/126 fathers in the FICare model (71%, mean age 35.1 (4.8) years, 98% male) and 93/137 (68%, 36.4 (5.5) years, 99% male) fathers in SNC completed questionnaires and were analyzed (see eAppendix 2). No differences were found in baseline characteristics between fathers who were responders and nonresponders (Appendix, eTable 3-4 in the Appendix). We included 3 same-sex partners, 2 in FICare and 1 in SNC. For baseline characteristics see Table 1. An imbalance in the gestational ages was present between the 2 groups, newborns in the FICare model had lower gestational ages (median $32^{+1}$ (IQR: $30^{+1}$ – $35^{+0}$) weeks versus $34^{+0}$ ($32^{+0}$ – $35^{+0}$) weeks, p=0.008, Mann-Whitney *U* Test) and longer hospital stays (median 39 (IQR: 15 – 58) days versus 21 (14 -36) days, p<0.001, Mann-Whitney *U* test). Fathers in the FICare group experienced a higher level of stress at birth than fathers in the SNC care group (mean 3.2 (SD: 1.3) versus 2.7 (1.2), p=0.03, students *t* test).

At discharge, 156/182 (86%) fathers completed questionnaires regarding their mental health and participation in newborn care during hospital stay (eTable 5-7 in the Appendix). At discharge, fathers' total stress score in the FICare model was lower than those of fathers in SNC units (adjusted beta -10.02, 95%CI -15.91 to -4.13, p=0.001, Table 2, eTable 8 in the Appendix). Fathers experienced less stress due to the environment and newborn behaviors in the FICare model (adjusted beta –5.748, 95%CI -10.14 to -1.356, p=0.01) compared to SNC. They also experienced less stress due to changes in their parental role in the FICare model (adjusted beta -4.271, 95%CI -6.536 to -2.006, p<0.001).

**Participation during hospital stay**

Fathers in the FICare model participated more in the care of their newborn compared with those in SNC (Table 2). Specifically, in the FICare model, fathers were more often able to be present and had higher total participation (adjusted beta 3.424, 95%CI 0.860 to 5.988, p=0.009). They searched less for information during hospital stay (CO-PARTNER tool *Domain* 3) and participated more in medical care (*Domain* 2, including tubefeeding, monitoring of the newborn, regulation of visitation to newborn and participating in daily rounds) than fathers in SNC. They also indicated being an advocate (*Domain* 4) of their newborn more. No differences were found for comforting of the newborn.

**Mediation analysis of parent participation on outcomes**

With mediation analyses we could distinguish the direct effect of the FICare model (through the *c'* path) and indirect effects through increased parent participation (the *ab* path). Two different scenarios arose from mediation analyses (Table 3).

1. *Beneficial outcomes associated with the FICare model that were explained by parent participation*

   Increased total participation in the FICare model was associated with fewer depressive symptoms (adjusted indirect effect -0.051, 95%CI -0.133 to -0.003) and lower impaired parent-newborn bonding scores (adjusted indirect effect -0.082, 95%CI -0.177 to -0.015) ((*ab* path), Table 3, Appendix eTable 9). No direct associations (*c'* path) for beneficial outcomes of the FICare model were observed for fathers' depressive symptomatology and parent-newborn bonding.

2. *Beneficial outcomes associated with the FICare model that could not be explained by parent participation*
   The FICare model was associated with less stress for fathers at discharge compared with fathers in SNC. Parent participation was not a mediator of this association (indirect effect 0.763, 95%CI -0.627 to 2.517). Fathers' participation in neonatal care was not a mediator of the association of the FICare model for fathers' self-efficacy at discharge (adjusted indirect effect 0.457, 95%CI -0.119 to 1.357) and also not for satisfaction with care (adjusted indirect effect 0.018, 95%CI -0.022 to 0.075).

# DISCUSSION

In this study in level-2 neonatal departments in the Netherlands, we found that fathers experienced benefits with implementing the FICare model in single family rooms with complete couplet-care for the mother-newborn dyad. In concordance with previous research we show that in our FICare model NICU-related stress in fathers was considerably lower[68], and we add to the literature with possible explanations through mediation analyses. The reduced stress is in line with associations of FICare in mothers[55] and single family rooms on mental well-being in fathers.[183] Despite baseline differences in gestational age of the newborns our results on mental health outcomes in fathers are in favor of the FICare model.

Our results suggest that it is especially the setting of the unit with single family rooms and complete couplet-care that supported fathers in diminishing stress. Interestingly, the reduced stress level was not explained by increased participation in care.

Fathers have to provide emotional support to the mother[441], manage the family's everyday life and may have to return to work quickly[441] during newborn hospitalization. They can perceive double burdens of concern for the well-being of the baby and the mother.[381] Also interpersonal factors such as beliefs regarding fatherhood[442], health care professionals' support[402] or parent-clinician communication[348,349] could potentially mediate the association between fathers' participation in care and stress. In addition, education and support to fathers might need to be different than the support to mothers, but preferentially qualitative research is needed to explore this more in depth.

We found positive associations of the FICare model for fathers' participation in care with depression and parent-newborn bonding. This finding complements previous literature[443] by showing that the ameliorated mental health of fathers of preterm newborns is mediated through parent participation.[196,444]

The FICare model in this study is a multi-component care model, that addresses parent-newborn separation and promotes parent participation through different aspects; namely the architectural design, integration of neonatal and maternity care, and a concomitant parent participation program. Solely addressing the architectural design does not improve mental health outcomes in parents and newborns.[68,147] Also, it is possible to participate in care in standard care settings, even without additional FICare. We addressed these issues with mediation analysis, discerning the associations of different aspects of parent participation (assessed by the CO-PARTER tool[207]) on fathers' mental health outcomes. This is important for current NICU care settings that are unable to change to single family rooms or couplet-care, as stimulating and endorsing parent participation can also be augmented in neonatal units with open bay settings.

Although we were unable to study the relation of the newborn to the father in this study, we believe that increased interaction in care and improved father-newborn bonding will also lead to a stronger reciprocal (emotional) relationship over time between father and newborn, which will be beneficial to the newborn as well.[445]

Our results suggest that fathers in the FICare model experienced less stress compared with fathers in SNC. Future research could include measurement of biomarkers (e.g. cortisol in hair or saliva) for better understanding of stress trajectories during newborn hospitalization and beyond.[212,446] Equally, universal screening of all expecting fathers and families on vulnerability for mental health issues (eg anxiety, depression and risk for impaired bonding) should be performed antenatally as part of routine care.

### Strengths and limitations

Strengths of this study include that we had a large sample of fathers. We included mediation analyses, to identify and explain the hypothesized association of increased parent participation in the FICare model with outcomes in fathers, with advanced statistical techniques[262] and a newly developed parent participation scale that was validated in fathers.[422] Also, fathers had high consent and response rates.

This study also has limitations. Most of the scales we used in this study were validated in women and mothers, in absence of suitable scales for fathers. As fathers too can feel depressed or anxious and have trouble coping with the birth of an ill or preterm newborn[447], future research should focus on developing and validating scales for fathers specifically. This will enable us to compare interventions across studies, but also to further support fathers in real time and according to their specific needs.

In absence of randomization due to hospital setting, we are unable to robustly confirm causality between participation and outcomes. Our results might also suggest a bidirectional association between participation and outcomes. For instance, fathers who were highly stressed participated more or fathers who were less depressed participated more. Therefore, future studies should incorporate randomization for instance on the hospital level ((stepped-wedge) cluster randomization) to evaluate hospital-based interventions.[448] However, with remodeling towards single family rooms and the complexity of NICU care culture, this might be difficult.

## CONCLUSION

In this study in level-2 neonatal units in the Netherlands, we found that an innovative FICare model with complete couplet-care for the mother-newborn dyad in single family rooms was associated with less perceived stress among fathers. In this FICare model, fathers can participate more, which is associated with fewer depressive symptoms and better parent-newborn bonding. Fathers should be enabled and supported to participate actively in all aspects of newborn care, and NICU care culture should be tailored to participation and the needs of fathers regardless of architectural design of the neonatal unit.

**Table 1. Baseline characteristics of fathers.**

| Characteristic | FICare group (n=89) | SNC group (n=93) | P-value |
|---|---|---|---|
| Age (mean (SD)) | 35.1 (4.8) | 36.4 (5.5) | 0.11 |
| Female (No (%)) | 2/84 (2%) | 1/86 (1%) | 0.62* |
| University degree (No (%)) | 74/82 (90%) | 75/85 (88%) | 0.87 |
| Paid job (No (%)) | 71/82 (87%) | 72/84 (86%) | 0.51 |
| Work hours per week (mean (SD)) | 39.7 (4.9) | 40.9 (7.6) | 0.27 |
| Identifies with Dutch cultural background (No (%)) | 75/84 (89%) | 71/86 (83%) | 0.30 |
| Stress of pregnancy (mean (SD), max score 5) | 2.1 (1.3) | 2.2 (1.4) | 0.53 |
| Stress of birth (mean (SD), max score 5) | 3.2 (1.3) | 2.7 (1.2) | 0.03 |
| Gestational age (median (IQR), range (min-max)) | $32^{+1}$ $(30^{+1} – 35^{+0})$ $(24^{+5} – 36^{+6})$ | $34^{+0}$ $(32^{+0} – 35^{+0})$ $(25^{+3} – 36^{+6})$ | 0.008 |
| Inborn newborn (No (%)) | 40/89 (45%) | 61/93 (66%) | 0.008 |
| Singleton pregnancy (No (%)) | 74/89 (83%) | 80/93 (86%) | 0.74 |
| First child upbringing (No (%)) | 61/83 (73%) | 57/85 (67%) | 0.46 |
| Plan for upbringing Together with partner (No (%)) | 83/83 (100%) | 80/83 (96%) | 0.25* |
| Smoking (No (%)) | 8/78 (10%) | 12/82 (15%) | 0.53 |
| Use of drugs (No (%)) | 4/78 (5%) | 2/80 (3%) | 0.44[a] |
| Use of psychotropic drugs (No (%)) | 0/87 | 1/92 (1%) | 0.11[a] |
| Alcohol use (No (%)) | 47/78 (60%) | 54/81 (67%) | 0.50 |
| Anxiety and depression at admission (median (IQR)) | 8 (3 – 14) | 5 (3 – 7.8) | 0.32 |
| Impaired parent-newborn bonding at admission (median (IQR)) | 9 (3 – 12.8) | 9 (8 -12) | 0.72 |
| Parent self-efficacy at admission (mean (SD)) | 60.4 (6.9) | 59 (5.9) | 0.43 |
| Stress at admission (mean (SD)) | 43.2 (20.1) | 41.5 (15.6) | 0.71 |

Data are n/N (%), mean (SD) or median (IQR). Denominators differ because of missing data. [a]: Fisher exact test, IQR: interquartile range, max: maximum, No: number, SD: standard deviation

**Table 2. Fathers' participation in neonatal care during hospital stay and mental health outcomes at discharge**

| | FICare (n=89) | SNC (n=93) | Beta | 95%CI | p-value | Adjusted beta[a] | 95%CI | p-value |
|---|---|---|---|---|---|---|---|---|
| *Participation in neonatal care during hospital stay* | | | | | | | | |
| Presence (hours/day, median (IQR)) | 8.9 (2.3 – 15.5) | 4 (2.1 – 5.9) | 0.531[b] | 0.268; 0.794 | <0.001 | 0.582[b] | 0.305; 0.859 | <0.001 |
| Presence (>8hrs per day, No (%)) | 47 (52.8) | 22 (23.7) | 3.675 | 1.793; 7.531 | <0.001 | 4.942 | 2.057; 11.88 | <0.001 |
| ***Total participation (mean (SD), max score 62)*** | 45.9 (8.0) | 43.6 (8.0) | 4.405 | 2.019; 6.792 | <0.001 | 3.424 | 0.860; 5.988 | 0.009 |
| **Domain 1** *Participation in daily care (mean (SD), max score 22)* | 16.2 (4.2) | 15.5 (4.2) | 1.369 | 0.061; 2.677 | 0.04 | 1.071 | -0.305; 2.446 | 0.13 |
| **Domain 2** *Participation in medical care (mean (SD), max score 8)* | 5.1 (1.9) | 4.5 (1.9) | 1.192 | 0.600; 1.785 | <0.001 | 0.861 | 0.264; 1.458 | 0.005 |
| **Domain 3** *Information gathering (mean (SD), max score 3)* | 2.3 (0.8) | 2.4 (0.8) | -0.159 | -0.418; 0.100 | 0.23 | -0.274 | -0.541; -0.008 | 0.04 |
| **Domain 4** *Advocacy and leadership (mean (SD), max score 3)* | 2.0 (1.0) | 1.7 (1.1) | 0.644 | 0.308; 0.980 | <0.001 | 0.518 | 0.162; 0.874 | 0.005 |
| **Domain 5** *Time spent with newborn (mean (SD), max score 12)* | 7.9 (2.9) | 7.2 (2.8) | 1.280 | 0.372; 2.187 | 0.006 | 1.464 | 0.463; 2.464 | 0.005 |
| **Domain 6** *Comforting the newborn (mean (SD), max score 14)* | 12.2 (2.0) | 12.2 (1.8) | 0.114 | -0.459; 0.687 | 0.69 | -0.176 | -0.790; 0.438 | 0.57 |
| *Mental health outcomes at discharge* | | | | | | | | |
| Stress PSS-NICU total score ( mean (SD)) | 40.8 (20.3) | 49.4 (18.9) | -8.589 | -14.56; -2.619 | 0.005 | -10.02 | -15.91; -4.130 | 0.001 |
| *Behaviour and sights and sounds* (mean (SD)) | 29.7 (14.1) | 34.8 (14.2) | -5.029 | -9.435; -0.623 | 0.026 | -5.748 | -10.14; -1.356 | 0.011 |
| *Parental role alteration* (mean (SD)) | 11.1 (7.5) | 14.6 (6.9) | -3.560 | -5.821; -1.299 | 0.002 | -4.271 | -6.536; -2.006 | <0.001 |
| Depression and anxiety (median (IQR)) | 7.0 (3.6 – 10.4) | 7.1 (3.3 – 10.9) | 0.065[b] | -0.143; 0.274 | 0.54 | 0.023[b] | -0.183; 0.230 | 0.83 |
| Self-efficacy score (mean (SD)) | 63.8 (6.9) | 62.2 (7.9) | 1.648 | -0.790; 4.086 | 0.18 | 1.459 | -1.100; 4.018 | 0.26 |
| Impaired parent-newborn bonding (median (IQR)) | 11.7 (5.1 – 18.1) | 9.4 (4.4 – 14.4) | 0.134[b] | -0.098; 0.367 | 0.26 | 0.137[b] | -0.109; 0.382 | 0.27 |
| Satisfaction with care (mean (SD)) | 5.2 (0.5) | 5.2 (0.6) | 0.055 | -0.111; 0.220 | 0.52 | 0.085 | -0.085; 0.255 | 0.32 |

Outcomes are from multiple imputed datasets. [a]: adjusted for gestational age, education, cultural background, age, stress at birth, work hours per week, upbringing plan, paternal smoking and alcohol use. [b]: after log transformation 95%CI: 95% confidence interval, FICare: family integrated care, IQR: interquartile range, n: number, SD: standard deviation, SNC: standard neonatal care

**Table 3. Mediation analysis of parent participation in neonatal care on outcomes at discharge**

| Outcome | Effect of the FICare model on mediator (participation) (*a* pathway (SE)) | Effect of mediator (participation) on outcome (*b* pathway (SE)) | Indirect effect (*ab*) | 95% CI of indirect effect (*ab*) | Effect of the FICare model on outcome (*c'*- pathway (SE)) | Effect of the FICare model on outcome (*c*-pathway (SE)) |
|---|---|---|---|---|---|---|
| *Crude analyses* | | | | | | |
| Self-efficacy | 4.405 (1.207) | 0.147 (0.074) | 0.649 | -0.068; 1.736 | 0.997 (1.289) | 1.648 (1.230) |
| Satisfaction with care | 4.405 (1.207) | 0.004 (0.006) | 0.018 | -0.0312 ; 0.082 | 0.037 (0.088) | 0.054 (0.084) |
| Depression and anxiety[a] | 4.405 (1.207) | -0.016 (0.007) | -0.069 | -0.155; -0.008 | 0.134 (0.109) | 0.065 (0.106) |
| Impaired parent-newborn bonding[a] | 4.405 (1.207) | -0.024 (0.007) | -0.107 | -0.206; -0.036 | 0.242 (0.118) | 0.134 (0.118) |
| Stress | 4.405 (1.207) | 0.255 (0.199) | 1.121 | -0.610 3.282 | -9.715 (3.152) | -8.589 (3.023) |
| *Adjusted analyses[b]* | | | | | | |
| Self-efficacy | 3.424 (1.295) | 0.133 (0.079) | 0.457 | -0.119; 1.357 | 0.999 (1.341) | 1.459 (1.292) |
| Satisfaction with care | 3.424 (1.295) | 0.005 (0.006) | 0.018 | -0.022; 0.075 | 0.067 (0.088) | 0.085 (0.086) |
| Depression and anxiety[a] | 3.424 (1.295) | -0.015 (0.007) | -0.051 | -0.133; -0.003 | 0.074 (0.107) | 0.023 (0.104) |
| Impaired parent-newborn bonding[a] | 3.424 (1.295) | -0.024 (0.008) | -0.082 | -0.177; -0.015 | 0.219 (0.122) | 0.137 (0.124) |
| Stress | 3.424 (1.295) | 0.223 (0.192) | 0.763 | -0.627; 2.517 | -10.78 (3.026) | -10.02 (2.977) |

[a]after In transformation, outcomes are from multiple imputed datasets, [b]adjusted for gestational age, education, cultural background, age, stress at birth, work hours per week, upbringing plan, paternal smoking and alcohol use

# Chapter 11

## General discussion

Overall, this thesis confirms and builds upon previous evidence, that including and accommodating parents as “new” and equal partners into neonatal care and research improves outcomes in families and their preterm infants who require hospitalization in a NICU.

Here, I will discuss the current perspectives on parent-infant closeness and parents as active partners in neonatal care. First, I will discuss the relationship between parent-infant closeness and parent partnered care models. Then, I will discuss the impact the configuration of the care environment has on morbidity and mortality in preterm infants and their parents. In addition, I will discuss the role of parent-provider communication and active parent participation and collaboration within the NICU context. Additionally, I will touch upon the facilitators and barriers health care professionals encounter to keep families close and participate in neonatal care. Next, I will propose my view on the way forward to implement parent-infant closeness and true partnership in neonatal care supported by research in an innovative FICare model. Lastly, I will give my perspective on future neonatal care improvement and research. The focus in this chapter is on health benefits for preterm infants and their parents, whenever we endorse parents as important contributors to the neonatal care team, with all team members providing their unique and skilled care for the infant.

## The relationship between parent-infant closeness and parent partnered models of care

Parent-infant separation during newborn hospitalization is still very common, which we confirmed in our study in NICUs throughout Europe **(Chapter 5).**[31,449] In European hospital settings (with a common (historical) division and compartmentalization between maternity and neonatal care) mothers and infants still need to be separated if one or both requires (highly specialized) care (42/45 (93%) of studied units).[449] More recently too, unfortunately, the COVID-19 pandemic highlighted the detrimental effects of non-participation of parents in neonatal care and separating parents from their newborns in NICUs, as was highlighted in a systematic review.[218] During this pandemic, families had restricted access to their loved ones, digital platforms were installed to replace personal contact and many NICUs restricted parental access to one parent (usually the mother). Depending on the region or clinical circumstances, they significantly restricted the amount of time parents could spend with their infant (sometimes as low as 5-15 minutes per day).[450,451] De-implementation of parent-partnered care models increased parent-infant separation, and parent disempowerment in infant care resulted in increased parental anxiety and depression. Parent-infant separation limited breastfeeding, increased parental stress, and decreased parent-infant bonding.[218,219,429,452–454]

The physical presence of parents in the NICU promotes their possibility to participate in (daily and medical) care, and parents can feel emotionally more connected to their infant as we also showed in our non-randomized trial (**Chapter 9 and 10**) and systematic review of the literature **(Chapter 4)**.[49,51,68,73,205,222] However, one should bear in mind, that if parents do not feel part of the NICU care team, or if their ability to make decisions is restricted, they can also feel (emotionally) separated from their infant, independent of the co-care facilities and infrastructure of the ward.[51] Equally, solely being physically present or supporting physical presence with the single family room design, does not necessarily imply that parents participate in care, are integrated in the neonatal care team or that they feel emotionally close and attached to their infant. Parents need to be part of the health care team and understand that they have an important role to play in neonatal care.[70] Parents can feel close to their infant when they believe they are enacting their parental roles, particularly when acting autonomously and making decisions about the care of their infant.[51] Contrarily, parents can experience trauma while their infant is hospitalized in the NICU and they cannot take on normal parenting roles and cannot be with their infant continuously or whenever they want.[49,59]

### Accommodating parents to be present within neonatal units results in beneficial outcomes in infants and parents

Accommodating parents to be present within NICUs, whenever they want, including during the night, should be strived for by hospitals and neonatal units. Previous studies have shown that design features of NICUs such as single family rooms, parent's bed next to the infant, bathroom and cooking facilities encourage and enable family presence.[65,187] Another study in Canada, suggested that the presence of a bed or couch signals the importance of parental presence.[455] If hospitals have the opportunity to change the design of the unit, this should be a joint effort of all stakeholders. By truly honoring the voices of parents to induce positive changes that matter to women, infants and families, one can successfully transition towards a single family room NICU design as was previously described by de Salaberry et al.[213] SFR designed NICUs are associated with better short and longterm healthcare outcomes compared to open bay unit settings as we showed in two systematic reviews and meta-analyses **(Chapter 2 and 4).**[68,456] The current literature on hospitalizing preterm infants in single family rooms contains no evidence on detrimental neurodevelopmental effects[456], one of the main discussion points previously suggested.[115] In contrast, as Lester et al. suggests in their study, an improvement in neurodevelopment can be mediated in the SFR by higher maternal involvement and developmental support in the SFR design.[134] Other results considering outcomes in infants hospitalized in a single family room setting, were that they experience less late-onset sepsis events **(Chapter 2 and 3)**[262,456], have higher chances of leaving the hospital at discharge exclusively breastfed **(Chapter 2)**[456] and experience less morbidity and mortality **(Chapter 2 and 3).**[262,456]

In **Chapter 3** we identified intravenous catheters and parenteral nutrition as a potential mediator for the beneficial association with decreased late-onset sepsis in the FICare setting with a SFR design.[262] The reason for the decreased use of catheters and parenteral nutrition remains to be further elucidated. One could hypothesize that the presence of parents, who know their infants well, may have resulted in less antibiotic treatment for symptoms and signs that were interpreted by medical caregivers as clinical symptoms for late onset sepsis.[457] It is interesting to note, that previously parents were seen as a potential source of infection for their infant, but these results in single family rooms support a hypothesis of a *protective* effect of parents, which early in the 1970s was already discussed by Barnett et al.[3,458] If parents are included in rounds, and their opinions are valued by staff, parents might aid in the treatment of their infants during hospital stay and reducing mortality and morbidity, with information that is very valuable and should be taken into account.[459] As an example, if parents can actively participate in rounds, they can provide medical professionals with additional medical information on the current state of their infant. Such as, the “overall well-being” of their infant (for example; “my daughter/son is feeling good today”) but they could also relay medical information (for example: “he/she had less apnea and bradycardia periods the past 24 hours, breathing is sometimes shallow”). Another hypothesis could be that, in the single family room design, adherence to hand hygiene is better[142] or that parents act as gatekeepers for medical personnel to enter or bundle interventions. It could also be, that by enabling parents to be present, this leads to better breastfeeding rates, and to different (protective) microbiota contamination of the single family room.[126] Human breastmilk is the optimal food for newborn infants, which amongst many, contains immunoglobulins and immunomodulatory components that can protect and treat infants from infection.[460]

### Measuring parent participation and collaboration in neonatal care

Up till recently no validated measure was able to capture the construct of parent participation and collaboration within the NICU context. Previous studies have shown that interventions that increase parent participation and skin-to-skin contact are beneficial in preterm and very low birth weight infants, however none directly recorded or measured this construct of participation and collaboration in their studies in the absence of a validated parent participation measurement tool.[34,55,94,95,112–114,116–118,121,131,134,135,181–187] With the development and psychometric evaluation of our CO-PARTNER tool, we now have a tool to measure collaboration on NICUs between parents and healthcare providers and active parent participation, also from the parents' perspectives **(Chapter 6).**[422] All items included in the CO-PARTNER tool can be performed by parents and this should be fully supported by units, as is advocated by parent representatives and the WHO.[78,261] This is different from other measures that have been previously reported, focusing on (the passive construct of) presence of parents using a diary[187] and health care professional evaluation of parent competencies and activities.[116,134]

With CO-PARTNER scores parents themselves can provide actionable quantitative data on the level of parent participation in care, with lower scores suggesting more tasks performed solely by health care professionals without participation of parents. It could identify gaps in proficiency or level of participation and collaboration of parents for instance in providing tube feeding. One could imagine, that if a parent is -hypothetically- not aware of the possibility to participate in tube feeding practices, by filling out the CO-PARTNER tool item on tube feeding, the parent could become aware that this is a part of neonatal care that they could actively participate in. Subsequently, the parent and nurse could discuss outcomes of the CO-PARTNER tool and make plans to improve the parent's participation in this item. It can therefore enable parents and professionals to openly discuss their partnership in real-time and to evaluate parent progression in independently taking care of the infant. At the unit-level it can be part of quality improvement cycles of continuously measuring the extent and quality of parent participation and collaboration in neonatal care. It could also help identify potential room for improvement of parent participation on the unit. Equally, the CO-PARTNER tool can enable comparison of parent-partnered care practices and to study (health) outcomes in infants and their parents through, for instance, mediation analysis (as we also showed in **Chapter 9 and 10**).[73,222,262] Currently, the tool is being translated into several languages (German, Chinese, Norwegian and Portuguese), making it more widely available and accessible.

### Appropriate language and communication is needed to achieve true partnerships in neonatal care and research

To achieve partnership in neonatal care, communication is pivotal and the vocabulary we use matter.[95] Words such as "visiting" and "allowing" degrade parents' role in the NICU and our communication can have (longstanding) effects on parents **(Chapter 8).**[461] Studies exploring parents' experiences of their time in the NICU confirm the crucial role of adequate verbal and non-verbal communication between parents and staff.[299,304,313,346–350] Parents value language that makes them feel emotionally supported, treated with empathy, and taken seriously.[278,294,299,348,349,351] They prefer clear, accurate, accessible, and timely information in which they are able to share their own knowledge.[289,314,348,349,352] Finally, both parents and providers emphasize the need to empower parents through training and positive feedback so they learn to care independently for their infants during admission and are well-prepared upon discharge.[290,292,308,322,462] Optimal parent-provider communication can build and maintain stable and equal relationships, enable information exchange, promote (sharing) decision-making, and enforce parent self-management **(Chapter 7 and 8).**[348,349] Adequate interaction entails that providers seek to consistently pay attention to the *topic*, *aims*, *location*, *route*, and *design* of their communication – thereby adapting their communication to parents' needs in a given situation. Providers may use the acronym TAILORED as a mnemonic to remember the five factors of adequate communication **(Chapter 7).**[349]

Through these functions, health care professionals can impact parents' coping (stress), knowledge, participation, parenting, and satisfaction **(Chapter 7 and 8).**[348,349] However, parents still report suboptimal communication and perceived gaps in communication [348] Not only being aware of the importance of communication in the NICU will aid in better communication strategies. Health care providers also need to be trained, as advocated by Benzies et al. on the importance of recurring healthcare education in relational communication, to optimize FICare practices and promote partnership in the NICU.[95,416,463] One of the points of attention are language barriers.[464] Language translation for improvement and extending family integrated care in the neonatal unit is needed[465], and pictorial support or interpreters could therefore be used.[396,466]

### Beneficial mental health outcomes in parents are associated with active parent partnership in neonatal care

In this thesis we studied the association between an innovative FICare model, active parental participation in neonatal care and maternal and paternal mental health outcomes **(Chapter 9 and 10)**.[73,222] Within a large teaching hospital with a level-2 neonatal unit in Amsterdam, the Netherlands, this innovative FICare model was set up as a multicomponent care model that addresses parent-newborn separation and promotes parent participation through different aspects, namely, the architectural design, integration of neonatal and maternity care (couplet-care in single family rooms), and a concomitant parent participation program based on the principles of FICare.[200] In couplet-care mothers and infants are cared for together, side by side. Within this innovative FICare model, the 4 C's of Closeness (Culture, Collaboration, Coaching, Capacities) are all addressed (**Chapter 5, 9 and 10)**.[73,222,449] Our studies show, that in this FICare model of care, stress due to specifically mother-infant separation is diminished **(Chapter 9).**[222] This is potentially partly due to the implementation of couplet-care, building upon previous work, that whenever co-care is provided to mothers and infants, this diminishes maternal feelings of stress.[67,118,213] However, as our studies also suggest, it might not solely be the care setting. Parent participation, operationalized in 6 domains (daily and medical care, advocacy, seeking information, time spent with the infant, closeness and comforting the infant, **Chapter 6)**[207] was a large mediator of the beneficial association of the FICare model and stress, self-efficacy, mother-infant bonding, and depression and anxiety (**Chapter 9)**.[222] In other words, if mothers can participate and collaborate more in neonatal care, and feel close to their infant, this is associated with ameliorated maternal mental health, which is in line with previous trials on the effect of FICare.[55]

### Never forget fathers

Up till now, scarce research has been conducted in fathers of neonates and the effect of parent partnered neonatal care models on their well-being. Fathers (and partners) can play an important role during infant hospital stay but they often feel excluded from their baby's caregiving and decision making, and can experience the double burden of concern for the well-being of the baby and that of the baby's mother.[381,467] They have to support mothers and want to participate in care of their infant, while they also have to manage the family's everyday life, traveling between hospital and home, and potentially also have to take care of other siblings.[441] They can experience trauma and depression following preterm birth[437,438] and experience uncertainty, distress, and a lack of control if they are not involved in their infants' care.[289] They too can experience surges in oxytocin release during skin-to-skin care and feelings of closeness and proximity just as mothers, but their paths towards these feelings of closeness might be different.[467–469] Evidence in this thesis suggests that in our facility with FICare and couplet-care for mothers and infants, fathers have less stress and are able to participate more in care which is associated with lower depressive symptomatology, and better parent-infant bonding scores than in standard care settings **(Chapter 10).**[73] This is additional to other publications, stressing the importance of paternal involvement in neonatal care, interacting with their infant, and tailoring NICU education and coaching to fathers specifically as well.[470,471]

### Parents can have an additional and unique role in neonatal care

Parents have their own competencies, are not trained as health care professionals, but need information and coaching during infant hospitalization to take on their preferred parental role.[199,463,472] They have an irreplaceable role within the health care team, that is additional to the professional specialties that are already present. For instance, as a recent systematic review and meta-analysis of randomized controlled trials highlighted, if interventions are applied that include direct parental bedside care in the NICU, this is associated with health benefits for the infant (increased weight gain velocity, decreased incidence or retinopathy of prematurity, decreased length of hospital stay, increased breastmilk intake, better neurodevelopment). Beneficial outcomes associated for the parents are also present, such as decreased parental stress and anxiety.[473]

Including parents as true partners in neonatal care and achieving zero-separation during hospital stay can be challenging **(Chapter 5).**[449] Currently, staffing shortages and high workloads are posing a serious threat on the application of parent-infant closeness and FICare as was also found as part of the theme *Capacities*.[449] This is especially true for the nursing work-force, hence retention and establishing a positive work environment for nurses is now potentially of even more importance.[474] Interestingly, in a study in the US, implementation of FICare was associated with increased nursing job-satisfaction without increasing work-load.[475]

Delivering FICare or other parent partnered models of care, can have influence on the requirements and identities of health care providers themselves, as was recently studied by Stelwagen et al. in the same innovative FICare model with single family rooms that we studied in **Chapter 9 and 10**.[463] The constant proximity of parents to their newborns, requires health care professionals to focus the attention on aligning the care of the newborn in full partnership with the parents. Health care professionals need training to become competent in the new way of working, and being able to coach and educate the parents, which is also part of the theme *Coaching* in our European study (**Chapter 5**).[449,463] Health care providers also need emotional support to be able to take care of these vulnerable families.[465] An excellent intervention that can be seen as example for other units, is the *Close Collaboration with Parents™* program, a comprehensive and systematic training program in which all staff are taught new skills to collaborate with parents. It entails several phases, and includes multifaceted mentoring on the bed-side and in discussions on the individual and unit level.[476] It aims and has shown to successfully promote a *Culture* shift in the unit and support parental presence and involvement in infant care.[53,54,476–478]

It seems to be a balancing act to offer staff enough resources, education and time to enable active parent participation in care of these vulnerable patients, which could be very much context specific and complex. It requires management and decision makers to support staff and to listen to frontline staff as to specific areas that need additional quality improvement efforts.[476] Parents and their experience and knowledge are often overlooked or not fully supported in (hectic daily) clinical care, research and in quality improvement endeavors. In one US study, family centred and integrated care practices were overall strongly endorsed, but there were indications of lack of knowledge or valuing of several key dimensions and gaps between current and necessary family care practices. This was especially true for family partnership at the organizational level.[465]

It is therefore important to not only focus on supporting parental presence or parental bedside care in the unit. As the Framework of Carman et al. poses, there are three levels of patient/family engagement: direct care, organizational design, and policy-making.[479,480] Parents should therefore also be given the possibility to be integrated into care delivery systems and be enabled to contribute to improving healthcare from the bedside to the health system boardrooms (for instance through patient and family advisory councils).[84,480,481] But also parent-led organizations can be of assistance. They can for example provide education for families and health care professionals. But they can also deliver mental health programs, organize peer support groups and can give training to hospitals to create peer support groups. By partnering with (veteran) parents or parent-led organizations, one can improve family care practices in the neonatal unit.

## Strengths and limitations of this thesis

This thesis encompasses several different study designs and views on parents as partners in neonatal care. We have performed systematic reviews **(Chapter 2, 4, 7 and 8)**[68,218,349,482,483], a qualitative study **(Chapter 5)**[73], a retrospective cohort study **(Chapter 3)**[262], a psychometric study **(Chapter 6)**[422] and prospective observational cohort studies **(Chapter 9 and 10).**[73,222] We also used an advanced statistical technique (multiple imputation by chained equations with mediation analyses and bootstrapping) for analyses to gain further insight into mediating factors that are enhanced by parent partnership in the neonatal unit **(Chapter 3, 9 and 10).**[73,222,262] Missing data are often a problem in epidemiological studies, with the potential to undermine the validity of results and have a potential to introduce bias.[158] By including multiple imputation by chained equations and performing sensitivity analyses based on missing data assumptions made in several studies (**Chapter 3, 6, 9 and 10)**[73,207,222,262] we have aimed to improve the presentation and soundness of our results.

Parents were active partners and played an active role in study designs (see "Patient Participation in this Thesis"), making this thesis clinically relevant to daily practice also from the parents' perspective. Above, we have included a rather large sample of fathers in our research, who are often overlooked in studies concerning this topic. We also set up a European network of neonatal departments, which has resulted in future collaborations on FICare.

However, this thesis should also be considered in light of its limitations. This thesis did not specifically incorporate health care professional views and effects of different intervention strategies, other than the challenges they face when implementing parent partnered neonatal care models and parent-infant closeness **(Chapter 5).**[484]

For instance, our systematic reviews showed beneficial outcomes for parents and their infants hospitalized in single family rooms **(Chapter 2 and 4)**.[68,485] However, especially for neonatal nurses, changing the architecture of the ward, requires them to work differently from how they are trained or are used to deliver care, while the evidence is still ambiguous on the impact on their psychological and physical wellbeing.[463,486–489] In some studies in the SFR design, neonatal nurses experienced better environmental quality, more patient-friendly care, more control of the primary workspace, more privacy and less interruption. But downsides such as feelings of isolation and increased walking distances have also been reported and need attention.[488,490–494]

In light of the impossibility of randomization due to architectural design, we performed several non-randomized studies. In our prospective non-randomized study (**Chapter 9 and 10**)[73,222], we had different enrollment numbers between the FICare model and standard settings. This was mainly due to non-consent in standard care, and not due to missed participants (these numbers were similar between settings), and therefore selection bias is not completely ruled out. It would therefore be interesting to study if there is an assocation with the care setting and consent to future studies. In our other non randomized before-after study (**Chapter 3**)[262], a possible explanation for a decreased incidence of sepsis and decreased use of catheters and parenteral nutrition could also be the effect of time itself. As we studied two different time periods (before and after start of the FICare setting), it might be possible that over time catheters or parenteral nutrition were used less or that health care professionals became more conservative in starting and/or more liberal in discontinuing antibiotics, which would be in concordance to previous studies.[177] Additionally, the potential causality which might be suggested with the mediation analyses that we performed **(Chapter 3, 9 and 10)**[73,222,262], should also be considered. For instance, mothers who are less depressed/better bonded/highly self-efficient might also participate more in care, and health care professionals should consider this when implementing programs aimed at increasing parent participation. Lastly, the included parents in our studies (**Chapter 6, 9, and 10)**[73,222,262] were overall highly educated and had a Dutch background. Therefore, the generalizability of some of the studies is to be taken into account, specifically since a high social economic status is for instance associated with more and longer exclusive breastfeeding.[495]

Most of the studies, were done within the context of the Netherlands in a level 2 neonatal unit. Outcomes for parents and their infants could be different in other settings, for instance in intensive care settings (level 3/4 NICU), or settings with acute medical care problems. In these "more intensive settings" implementing FICare can be very challenging, also considering the fragility of the population. However, recent studies have been suggesting to recommend skin-to-skin contact as a place of care, rather than a care intervention itself.[496,497] Therefore, this could also be a starting point for extending FICare to the very preterm population.

## Future perspectives on partnership in neonatal care and research

### Changing hospital neonatal care culture

To address issues concerning parent-infant separation, to ameliorate parent partnered neonatal care models and to further improve parent and infant outcomes, healthcare systems and all stakeholders need to prepare for change and have open mindsets.[223,498] This might be accomplished by creating stakeholder networks and learning platforms[499], embedding parents throughout all the layers of organization and by sharing stories of success. Ultimately, we need to aim for a *Culture* shift in neonatal care in which parents can be recognized as knowledgeable and irreplaceable NICU team members **(Chapter 5)**.[215,221,484] However, changing hospital care culture can be challenging and very difficult[221], but systematic principles to change hospital care culture have been previously proposed by Braithwaite et al.[214] I would like to build upon, translate and adopt these principles to the NICU-context.

One can start to improve parent partnered neonatal care models by using widely available Quality Improvement (QI) models, frameworks and techniques that have previously proven to advance NICU clinical care and outcomes such as infection prevention and ventilator management.[500–504] At the core for these QI to be successful, one needs to build a cohesive team that includes former NICU families.[505] As stated before, parents should be recognized as knowledgeable and irreplaceable team members and they should be facilitated, educated and supported to share their knowledge and experience. With this approach, one can potentially reciprocally improve neonatal care within the units from a parent-partnered perspective and improve parent-partnered neonatal care itself. One can use resources such as the Agency for Healthcare Quality evidence based guidelines toolkit for QI partnership among patients, families and health professionals.[506]

Then, attention must be given to how care is delivered at the cotside. To address how care is delivered at the cotside, measurement and ongoing monitoring are essential components of any quality improvement program. To evaluate parent partnered care models, units should invest in quality and qualitative evaluation concerning (the level of) parent participation and partnership in their unit (for instance by using the CO-PARTNER tool **(Chapter 6)**[207]**)** and implementing recurring FICare education for (newly aspiring) health care professionals.[422,507] Second, all meaningful improvement should be local and tailored to the specific NICU setting, there is no one-size fits all. Thirdly, daily successes on the unit are also present, and we need to appreciate and acknowledge how NICU professionals handle dynamic and complex situations and identify factors for success. Previous research[508] has shown that success stories usually have four common factors: begin with small scale initiatives and build up; convert data and information into intelligence and give this openly to the appropriate decision makers; remember that the lone hero model does not work and that collaboration underpins all productive change; and always start with the patient at the center of any reform measure.[214,508]

**Future research**

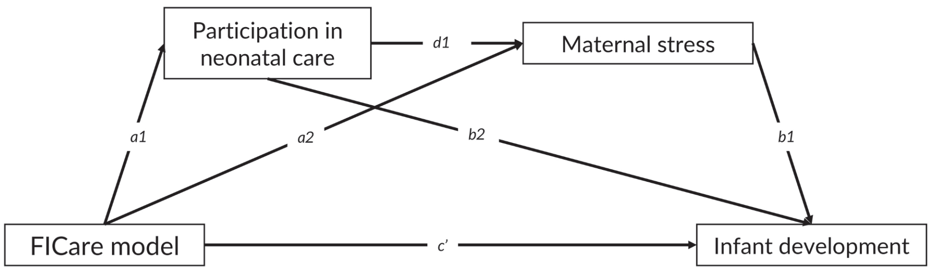
Partnership and encounters between professionals and parents (for instance communication strategies) could be addressed more thoroughly in future research. For the future I foresee the implementation and study of large datasets and the creation of decision trees or prediction models. For instance, discharge or transfer to a different hospital are often stressful periods during infant hospitalization and many parents feel unprepared when this happens, but also healthcare providers may have trouble planning this.[509] It would be interesting to study if data can be extracted from medical records and used to predict and anticipate hospital discharge, as is currently being studied in adult populations.[510] Prediction models could help parents in their neonatal trajectory, potentially reduce length of stay or parents' stress, and aid in the parent-provider communication on the neonatal unit.[389,509,511] Also, virtual reality or video simulations might be an interesting mode to educate, train and study staff in parent partnership and communication.

We need robust research designs to study associations and causality within neonatal care, especially when studying parent partnership and hospital care culture change. Future studies should be preferably randomized. However, with hospital-level interventions, and changes in unit culture, randomization is difficult on the individual patient level, with the potential for cross-contamination. Therefore (stepped wedge) cluster randomized trials should be performed with meaningful outcomes. Above, to gain in-depth insight in processes and changes, one always needs to consider conducting qualitative studies as these can be very meaningful and complementary to quantitative studies. For example, it would be very important to study settings that encounter many barriers to change towards more parent partnership or FICare practices, to understand their challenges and enable specific interventions to ameliorate newborn care.

As it is becoming clearer that fathers (partners) too can feel depressed and have trouble coping with the birth of an ill or preterm infant[447], future research should focus on developing and validating questionnaires for fathers specifically. This will enable us to compare interventions across studies, but also to further support fathers in real-time and according to their specific needs.[512] FICare programs should try to be tailored to the needs of fathers specifically, and we should never forget fathers while caring for the mother and her infant.

The outcomes of the preterm infants included in the AMICA study[236], and especially their neurodevelopment after admission to the innovative FICare model, are currently being evaluated, for the short (18-24 months age) and longer term (5 years of age). In following to previous research, it might be expected, that FICare will ameliorate outcomes in these infants too, in addition to previous research on extremely preterm infants studied in level 3/4 units.[513–515] Previously it has been shown that enrollment in FICare leads to lower child internalizing and externalizing behavior through improvements (mediation) of maternal cortisol, which is indicative of maternal stress levels.[514] It would be interesting to study, if the innovative FICare model, potentially has a cumulative/serial mediation effect on child outcomes, and if it affects outcomes relevant to the preterm infants in adult life and their quality of life. One could hypothesize for instance, that through increased parent participation (as measured with the CO-PARTNER tool, **Chapter 6**), maternal stress is decreased (**Chapter 9)** resulting in improved infant development (see Figure 1). This would be in line with the findings from an observational cohort study, finding that high stress, depression and anxiety scores in mothers were associated with an increased risk of communication delay and personal-social delay at three years of age in infants.[516] But also, if parents are true partners during infant hospitalization in the NICU and are able to participate in neonatal care, they could reduce and mitigate pain experiences of the infant during hospitalization.[517] Following this, one could hypothesize, that also physical health could be impacted on the longer term because prolonged and unmanaged painful procedures during NICU stay are associated with chronic pain that continues into adulthood, longterm changes to stress response and brain volume and development.[22,517] Other pathways might also be present, and this is in need of future research, and could for instance be studied with serial mediation analysis.

**Figure 1. Serial mediation analysis**

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Previous studies have shown, that neonatal intensive care is cost-effective, but the economic impact of FICare and its associated ameliorated outcomes in the short and longer term for the infant and family remains to be elucidated. For example, cost-reducing interventions that could be associated with implementation of FICare are transfer of expensive hospital care to home earlier and more structurally (for instance tubefeeding and cardiorespiratory monitoring) with telemedicine, as parents in FICare units are better and earlier prepared to take care of their infant independently.[518] One study in Canada has yet to publish their results and also our stepped-wedge cluster randomized trial in the Netherlands will look at this outcome.[95,96] It is important to study the association between FICare and the cost per quality adjusted life year and costs to society, as it is expected that these will be ameliorated and also other stakeholders such as unit management can be motivated to change hospital care culture.[476,519–521] Other potential relevant psychosocial and societal outcomes that could be impacted by FICare by improving the ability of the infant to form social relationships and bonds, could be romantic partnership and parenthood during adult life.[520]

Lastly, core outcome sets for neonatology have been defined[522] and future research should incorporate these outcomes in their studies. However, core outcome sets for parents of NICU infants are still lacking and in need of determination and definition. This will enable benchmarking, conducting systematic reviews and humanizing and ameliorating current neonatal practices more thoroughly. To define these core outcome sets several steps should be undertaken as outlined in Table 1 (adapted from[522–526]). In this process the input of all stakeholders, and specifically former NICU families, will be very valuable to further ameliorate neonatal care and research practices.

**Table 1 Development of a core outcome set for family integrated care practices**

| Step | Explanation |
|---|---|
| **1 – Systematic review** | Identifying outcomes reported in published studies regarding family integrated care |
| **2 – Qualitative phase** | Identifying outcomes important to all relevant stakeholders, not identified by step 1. |
| **3 – The Delphi process** | Determining which outcomes to include and exclude based on stakeholder input |
| **4 – Consensus meeting** | Arriving at consensus on outcomes to be included in the final core outcome set |
| **5 – Determination of outcome definitions and measurements** | Determining the most appropriate outcome definitions and measuring tools using a combination of systematic reviews and consensus methods |

## Conclusions

The current dogmas that underly parent-infant separation and insufficient parent participation in the NICU context should be addressed with multidimensional and evidence-based family care, research and quality improvement practices. Parents are one of the core pillars of a powerfully built construction in neonatal care. Having them as active and equal partners results in better infant and parent health outcomes, in the short and longer term. In order to achieve true partnership with parents and improve outcomes during and after hospital stay of their preterm infant, we need to involve parents and regard them as active, equal and irreplaceable partners in research and in care.

Chapter 12

# Summary
# Nederlandse samenvatting

# SUMMARY

In **Chapter 1** we describe the background and aims of the studies presented in this thesis. It describes the burden of prematurity (infants born before 37 weeks of gestation) and the problems parents encounter whenever their infant needs hospitalisation in the hospital on the neonatal (intensive) care unit. We provide an introduction on parent-partnered neonatal care models, parent infant closeness, and family integrated care as an extension of the care-by-parent program with 24 hour care by the mother, minimal use of technology and sparse contact between the baby and medical staff.

In **Chapter 2** we show with a systematic review and meta-analysis of the evidence that hospitalising preterm infants in single family rooms (SFR) compared to open bay units (OBU) is associated with health benefits. Thirteen study populations (n= 4,793 preterm infants) were included. Firstly, preterm infant hospitalization in a SFR environment does not lead to impaired neurodevelopment (n= 680 patients, MD+1·04, 95%CI -3·45, 5·52, $I^2$= 42%, p= 0·65), one of the main counterarguments previously given. However, most of these studies were performed in a subgroup of very preterm infants (born before <28 weeks of gestation). Secondly, preterm infants suffered from less infections in the SFR environment (approximately 37% less infections, n= 4,165 and 108,305 hospitalisation days, RR= 0·63, 95%CI = 0·50–0·78, $I^2$= 0%, p<0·0001). Infections decreased with 1 sepsis event per 1000 patient hospital days in infants admitted to SFR. Lastly, breastfeeding rates at discharge was higher in the group of infants hospitalized in SFRs compared with OBUs (n= 484, RR= 1·31, 95%CI = 1·07–1·61, $I^2$= 0%, p= 0·01).

In **Chapter 3** we present a retrospective before-after cohort study with 1,046 infants admitted to a level 2 neonatal unit, in Amsterdam the Netherlands. This unit in Amsterdam was rebuilt in 2014 from an open bay unit design towards a single family room design with complete couplet-care for the mother-infant dyad with family integrated care (FICare setting). Within family integrated care parents were trained to be the primary caregiver of their infant, and nurses supported, instructed and coached parents. Parents were invited but not obligated to be present $^{3}$8 hours per day, and rooming-in facilities were present if they wanted to stay during the night. Parents were actively encouraged to participate in their infant's care by providing feedings by nasogastric tube, breast or bottle, providing skin-to-skin care (by mothers and fathers), weighing and regulating temperature control. Family-centered rounds were implemented and included active parental participation in medical decision making on daily medical rounds and involving them in the process of patient management together with the nurses and doctors. Weekly, parents had group-sessions to learn and talk about prematurity and their infant's hospital stay, guided by health care professionals or veteran parents. Nurses provided cardiorespiratory monitoring as well as treatments such as intravenous fluids or antibiotics, placing nasogastric tubes, providing respiratory support and phototherapy.

In this before-after study, we studied 1,046 infants (468 in SFR (January 2015-December 2016), 578 in OBU (January 2012- June 2014), median gestational age 35 weeks). Infants admitted to the FICare setting suffered from less sepsis events (5.3% versus 9.3%) and could be discharged to home earlier than infants admitted to the standard care setting with open bay units (median length of stay 10 days versus 12 days). With mediation analyses, we were able to distinguish that the reduction in late-onset sepsis was mainly mediated by a reduced use of intravenous catheters and parenteral nutrition in the FICare setting.

After the results in infants, we focused on experiences of parents in neonatal care. We performed several studies in parents to elucidate burdens and health benefits of family integrated care approaches in neonatal care.

In **Chapter 4** we systematically reviewed the evidence on reconstruction of neonatal care units towards the single family room design, and the effect for parents of preterm infants. We included 11 study populations (1850 preterm infants, 1549 mothers and 379 fathers). Parents with infants admitted to SFRs had higher levels of parental presence, involvement and skin-to-skin care. At discharge, these parents had lower stress levels (n=828 parents, SMD-0·30,95%CI -0·50;-0·09,p<0·004,$I^2$=46%), specifically NICU-related stress (n=573, SMD-0·42,95%CI -0·61;-0·23,p<0·0001,$I^2$=0%). Parents also had higher levels of empowerment, perceived the unit to be more family-centred and had higher satisfaction levels. We did not find differences for anxiety, parent-infant bonding, self-efficacy, or depression. Parent-participation in care was assessed very heterogeneously in absence of a validated parent-participation measurement tool.

In **Chapter 5** we performed a qualitative study in 45 units throughout 18 countries in Europe and Canada. In this study we asked health care professionals on the setting and parent-partnered care practices they performed in the unit, and which burdens and facilitators they encountered to keep families close during maternity and/or neonatal care. We integrated parents in our research team, and they were part of all phases of study. Results showed that despite implementation of components of parent-partnered care models, parent-infant separation during infant and/or maternity care was very common (n=42/45 units, 93%) and limited parent-infant closeness and further upscaling of optimal parent-partnered care practices in units. NICU professionals encountered four main themes with facilitators and barriers on and between the hospital, unit, staff and family-level: *Culture* (jointly held characteristics, values, thinking and behaviours about parental presence on the unit), *Collaboration* (the act of working together between and within different levels), *Capacities* (resources and policies), and *Coaching* (education to acquire and transfer knowledge and skills).

In following to **Chapter 4**, we report in **Chapter 6** on the development and psychometric properties of an active parent participation tool (CO-PARTNER tool) for use in neonatal care. Items were generated in focus group discussions and in-depth interviews with professionals and (veteran) parents. The tool measures active parent participation and collaboration in neonatal care within six domains: *Daily Care*, *Medical Care*, *Acquiring Information*, *Parent Advocacy*, *Time Spent with Infant* and *Closeness and comforting the infant*. The tool was completed at NICU-discharge by 306 parents (174 mothers and 132 fathers) of preterm infants. Subsequently, we studied structural validity with confirmatory factor analysis, construct validity, using the Average Variance Extracted and Heterotrait-Monotrait ratio of correlations, and hypothesis testing with correlations and univariate linear regression. For internal consistency we calculated composite reliability (CR). We performed multiple imputations by chained equations for missing data. Analyses showed that the tool explicitly measures parents' participation and partnership with professionals in neonatal care incorporating their unique roles in care provision, leadership, and connection to their infant. The tool consists of 31 items within six domains with good face, content, construct and structural validity. This tool is able support parents and to study the effects and implementation of parent partnered models of neonatal care.

In the neonatal (intensive) care unit (NICU), parents continually engage in communication with healthcare staff. Communication between parents and providers serves important clinical goals; through interaction important information is relayed, consent is obtained, and decisions are made. In addition, communication can foster collaboration between parents and staff. In **Chapter 7 and 8** we synthesize and analyse the literature on the function and effects of parent-provider communication during infant hospitalization in the NICU on parent-related outcomes, introducing the NICU Communication Framework. We performed 2 systematic reviews of the published literature, including 43 en 77 publications respectively. NICU communication has four distinct, main *functions* of parent-provider interaction: building/maintaining relationships, exchanging information, (sharing) decision-making, and enabling parent self-management. Adequate interaction entails that providers seek to consistently pay attention to the *topic*, *aims*, *location*, *route*, *and design* of their communication – thereby adapting their communication to parents' needs in a given situation. Providers may use the acronym TAILORED as a mnemonic to remember the five factors of adequate communication. Across these functions, we found that parent-provider interaction has the potential to affect parent's: (1) coping, (2) knowledge, (3) participation, (4) parenting, and (5) satisfaction.

In **Chapter 9 and Chapter 10** we aimed to study the effect of a zero-separation model of neonatal care in Amsterdam the Netherlands on mental health and participation of mothers and fathers of preterm infants. In this model of care family integrated care in single family rooms with complete couplet-care for the mother-infant dyad (FICare setting) is given. We compared the FICare setting to separate maternity care with standard neonatal care in open bay units (standard setting). In a prospective non-randomised clinical trial (May 2017-January 2020, AMICA-study[236]) we included 296 mothers and 263 fathers of 358 preterm infants (median gestational age at birth 33+2). In **Chapter 9** we show that mothers of preterm infants in the innovative FICare setting had lower total stress scores at discharge than mothers in SNC. The FICare-setting was specifically associated with less stress due to mother-infant separation. Active participation in neonatal care was a partial mediator of the effect of FICare on stress and specifically participation in daily care, medical care, comforting and closeness with the infant and time spent with the infant. Participation, but not care setting, was a large mediator for less depression and anxiety, higher self-efficacy and bonding scores in mothers of preterm infants. Likewise, in **Chapter 10** we show that fathers in FICare also experienced less stress and had higher participation scores than in SNC. Participation, but not care setting, explained the effect of FICare on fathers' depression and bonding with his infant. The zero-separation model of neonatal care is therefore associated with improved outcomes in mothers and fathers of preterm infants, specifically considering stress.

The last chapter (**Chapter 11**) summarizes and discusses the main findings of our studies. It is followed by a general discussion of the relationship between parent-infant closeness and parent partnered care models. Additionally, the facilitators and barriers health care professionals encounter to keep families close and participate in neonatal care are discussed and the role of parent-provider communication within the NICU context. It also proposes a way forward to implement zero-separation and true partnership in neonatal care, how we can measure this, supported by research in our innovative FICare model. It eventually gives a perspective on future neonatal care improvement and research. The focus in this chapter is on health benefits for preterm infants and their parents, whenever we endorse parents as important contributors to the neonatal care team -with all team members providing their unique and skilled care for the patient- and limit parent-infant separation.

# NEDERLANDSE SAMENVATTING

**Hoofdstuk 1** bevat de achtergrond en onderzoeksdoelen van de studies in dit proefschrift. Het introduceert de ziektelast van prematuriteit (kinderen geboren voor 37 weken zwangerschapsduur) en welke problemen ouders van deze kinderen kunnen ervaren. Daarnaast beschrijft het "parent-partnered" neonatale zorg modellen, ouder-kind nabijheid (parent-infant closeness), familie-kamers (single family rooms) en familie-geïntegreerde zorg (Family Integrated Care) zoals dat een uitweiding is van het "zorg-door-ouder programma met 24 uur zorg door de moeder, minimaal gebruik van technologie en weinig contact tussen de baby en het medisch personeel, zoals dat voor het eerst was voorgesteld door Levin et al.[85]

**Hoofdstuk 2** beschrijft een systematische review en meta-analyse van de gepubliceerde literatuur. Het laat zien, dat wanneer prematuur geboren kinderen worden opgenomen op een neonatalogie afdeling met familie-kamers in plaats van op een zaal-afdeling dit geassocieerd is met betere gezondheidsuitkomsten. Hiervoor werden 13 publicaties met in total 4793 prematuur geboren kinderen opgenomen op een neonatologie afdeling geevalueerd. Een van de belangrijkste bevindingen is, dat wanneer kinderen worden opgenomen in een familie-kamer dit niet geassocieerd is met een slechtere neurologische uitkomst (n= 680 patiënten, MD+1·04, 95%CI -3·45, 5·52, $I^2$= 42%, p= 0·65). Dit was een van de belangrijkste argumenten tégen het implementeren van familie-kamers op de neonatologie. Echter, waren deze geincludeerde studies alleen uitgevoerd in een subgroep van extreem vroeg-geboren kinderen (geboren <28 weken zwangerschapsduur). Ten tweede blijkt, dat kinderen minder infecties doormaakten (ongeveer 37% mindere infecties, n= 4,165 patiënten 108,305 opname dagen, RR= 0·63, 95%CI = 0·50–0·78, $I^2$= 0%, p<0·0001) in familie-kamers. Gerekend in opname dagen, namen de infecties af met ongeveer 1 sepsis (bloedvergiftiging) per 1000 opname dagen bij kinderen opgenomen in familie-kamers. Tenslotte waren de percentages rondom borstvoeding positiever in prematuur geboren kinderen opgenomen familie-kamers vergeleken met prematuur geboren kinderen opgenomen op een zaalafdeling (n= 484, RR= 1·31, 95%CI = 1·07–1·61, $I^2$= 0%, p= 0·01).

**Hoofdstuk 3** presenteert een retrospectieve voor-na cohort studie met 1,046 opgenomen premature kinderen in een level 2 neonatologie afdeling (het Anna Paviljoen) in OLVG, Amsterdam, Nederland. Deze neonatologie afdeling was in 2014 verbouwd van een zaalafdeling naar een afdeling met familie-kamers met complete moeder-kind zorg (couplet-care) en familie-geïntegreerde zorg (family integrated care) (FICare setting). Met het toepassen van het familie-geïntegreerde zorg concept worden ouders getraind om de primaire zorgverlener te worden van hun kind, en ondersteunen, instrueren en coachen de verpleegkundigen vooral de ouders. Ouders worden uitgenodigd, maar niet verplicht, om $^{3}$8 uur per dag aanwezig te zijn op de afdeling, en faciliteiten voor 24-uurs aanwezigheid zijn beschikbaar voor ouders als ze ook willen blijven slapen. Ouders worden actief ondersteund om te participeren in de zorg voor hun kind zoals bijvoorbeeld het geven van sondevoeding, borstvoeding of flesvoeding, huid-op-huid contact (zowel door moeders als door vaders), wegen en temperatuur regulatie van hun kind. Tijdens de opname doen ouders mee in de dagelijkse visite, en kunnen ze actief meedoen in gedeelde besluitvorming (*shared-decision making*) en participeren ze in patiënt management samen met de verpleegkundigen en artsen. Ouders hadden wekelijks groeps-sessies met andere ouders om te leren en te praten over prematuriteit en het verblijf van hun kind. Deze sessies werden begeleid door professionals of door ouders die een eerdere ervaring hadden met een kind op een afdeling neonatologie afdeling (*veteran parents*). Verpleegkundigen concentreerden zich in deze setting op het geven van cardiorespiratoire ondersteuning en monitoring, het geven van (medische) behandelingen zoals medicijnen via het infuus of antibiotica, het plaatsen van de sonde, of bijvoorbeeld fototherapie.

In deze studie vergeleken we prematuur geboren kinderen (mediane zwangerschapsduur 35 weken) opgenomen in OLVG op de FICare setting (van Januari 2015 – December 2016, 468 kinderen) met de kinderen die daarvoor op de oude zaalafdeling opgenomen lagen (van Januari 2012-Juni 2014, 568 kinderen). Kinderen die waren opgenomen op de FICare afdeling hadden minder vaak een sepsis (bloedvergiftiging, 5.3% van de kinderen versus 9.3% van de kinderen). Het verminderde gebruik van infusen en specifiek parenterale voeding in de FICare setting was een belangrijke mediator voor deze mogelijke vermindering in het voorkomen van sepsis in de FICare setting. Tevens waren kinderen vaak minder lang opgenomen in de FICare setting (mediane opnameduur van 10 dagen) dan op de standaard zorg afdeling (mediane opnameduur van 12 dagen).

Na de uitkomsten in kinderen, focusten beschrijven de volgende hoofdstukken de uitkomsten en ervaringen in de ouders van de opgenomen kinderen. Hiervoor werden verschillende soorten studies uitgevoerd in ouders, om te achterhalen wat de gezondheidsvoordelen zijn van een familie-geïntegreerde neonatale zorg benadering.



**Hoofdstuk 4** beschrijft een systematische review en meta-analyse van de gepubliceerde literatuur, waarin de uitkomsten in ouders worden vergeleken wanneer hun prematuur geboren kind wordt opgenomen op een neonatalogie afdeling met familie-kamers met een zaal-afdeling. Uitkomsten van dit review laat zien dat dit geassocieerd is met betere gezondheidsuitkomsten in de ouders. Hiervoor analyseeerden we in totaal

11 populaties (1850 prematuur geboren kinderen, 1549 moeders en 379 vaders). Ouders met kinderen opgenomen op een afdeling met familie-kamers waren vaker aanwezig, waren meer betrokken bij de zorg en hadden meer huid-op-huid contact met hun kind. Bij ontslag van hun kind uit het ziekenhuis, hadden ouders lagere stress levels (n=828 ouders, SMD-0·30,95%CI -0·50;-0·09,p<0·004,$I^2$=46%), specifiek minder NICU-gerelateerde stress (n=573, SMD-0·42,95%CI -0·61;-0·23,p<0·0001,$I^2$=0%). Ouders hadden ook hogere levels van empowerment, vonden dat de afdeling meer familie-gericht was en waren meer tevreden met de zorg voor hun kind. Er werd geen verschil gevonden in de vergelijking voor angst, ouder-kind hechting, zelfredzaamheid, of depressie. Daarnaast bleek dat ouder-participatie in de zorg zeer heterogeen werd gemeten, omdat er geen gevalideerd meet-instrument voor is.

**Hoofdstuk 5** bevat een kwalitatieve studie op 45 neonatologie afdelingen door 18 landen in Europa en Canada. In deze studie werd onderzocht hoe de zorg voor moeder en kind werd vormgegeven en welke “parent-partnered” modellen van neonatale zorg er werd toegepast op de verschillende afdelingen. Daarnaast beschrijft het welke barrières en ondersteuning zorg-professionals ondervonden om families nabij te houden (parent-infant closeness) ten tijde van zorg voor de moeder en/of het kind. Ouders waren onderdeel van het onderzoeksteam, en ze droegen bij in alle fasen van de studie. Een belangrijk resultaat van deze studie was, dat ondanks dat er (componenten van) “parent-partnered” zorg modellen werden geïmplementeerd op afdelingen, moeder en kind nog vaak van elkaar gescheiden moeten worden (42/45 afdelingen, 93%) wanneer moeder en/of het kind zorg nodig hadden. Hierdoor konden ouders niet altijd optimaal nabij hun kind zijn, en konden parent-partnered modellen van neonatale zorg ook niet verder uitgebreid worden. NICU zorgverleners ondervonden successen maar ook barrières voor de implementatie van familie-nabijheid op 4 thema’s op het ziekenhuis-, unit-, personeels- en familie-niveau: *Cultuur* (gezamenlijke waarden, gedrag en denkwijzen over de aanwezigheid van ouders op de neonatologie afdeling), *Samenwerking* (samenwerken tussen en binnen de verschillende niveaus), *Capaciteiten* (faciliteiten en richtlijnen) en *Coaching* (scholing, zowel educatie en vaardigheden krijgen als ook overbrengen).

Volgend op **Hoofdstuk 4**, rapporteert **Hoofdstuk 6** over de ontwikkeling en validatie van een actieve ouder participatie vragenlijst (CO-PARTNER) voor gebruik in de neonatale zorg. Items werden gegenereerd in focus groep discussies en diepte-interviews met ouders met een prematuur geboren kind op een neonatologie afdeling en zorgprofessionals die werkten op een neonatologie afdeling. Met de vragenlijst kan men actieve participatie op een afdeling neonatologie meten binnen 6 domeinen: *Dagelijkse zorg*, *Medische zorg*, *Informatie verzamelen*, *Opkomen voor het* *kind*, *Tijd doorbrengen met het kind* en *Nabijheid en troosten van het kind*. De vragenlijst werd ingevuld bij ontslag van een prematuur geboren kind door 306 ouders (174 moeders en 132 vaders). Vervolgens werd de validiteit van de vragenlijst, construct validiteit getest en ook een aantal hypotheses met correlaties en univariate lineaire regressies. Tevens beschrijft de studie de interne consistentie van de vragenlijst. In de studie werden uitgebreide statistische analyses uitgevoerd om om te gaan met missende data (multiple imputation with chained equations). De analyses laten zien dat met de vragenlijst expliciteit ouders actieve participatie en samenwerking met zorgprofessionals wordt gemeten, in de unieke rol die ouders voor hun kind op een neonatologie afdeling hebben. De vragenlijst bestaat uit 31 items in 6 domeinen en heeft een goede validiteit. Daarnaast kan deze vragenlijst ook mogelijk ouders ondersteunen ten tijde van een opname op een afdeling neonatologie en helpen bij de implementatie en het meten van parent-partnered care modellen van neonatale zorg.

Op een neonatologie afdeling, nemen ouders continue deel aan communicatie met zorg-professionals. Communicatie tussen ouders en zorgprofessionals kan belangrijke klinische doelstellingen hebben: door interactie kan belangrijke informatie worden overbracht, kan toestemming verkregen worden en kunnen beslissingen genomen worden. Bovendien kan communicatie de samenwerking tussen ouders en zorgprofessionals ondersteunen. **Hoofdstuk 7 en 8** bevat een beschrijving van de synthesering en analyse van de literatuur ten aanzien van de functies en effecten van ouder-zorgprofessional communicatie ten tijde van een opname van een kind op een neonatologie afdeling. Dit werd gedaan met 2 systematische reviews van de gepubliceerde literatuur, waarbij er respectievelijk 43 en 77 publicaties werden geïncludeerd. De focus in de resultaten lag daarbij op de ouder-uitkomsten, en de introductie van het "NICU Communicatie Framework". Communicatie op een neonatologie afdeling heeft 4 afzonderlijke en hoofd *functies* wanneer men kijkt naar ouder-zorgprofessional interactie: 1. bouwen en onderhouden van relaties, 2. uitwisselen van informatie, 3. (gedeelde) besluitvorming en 4. ondersteunen van zelfredzaamheid (**Hoofdstuk 7).** Over de 4 verschillende functies heeft communicatie meerdere effecten op: (1) copings-mechanisme van ouders, (2) kennis van ouders, (3) participatie van ouders, (4) ouderschap, en (5) tevredenheid van ouders **(Hoofdstuk 8).** Adequate communicatie door zorgprofessionals kan worden bereikt als zij consistent aandacht besteden *topic* (onderwerp), *aims* (doelen), *location* (locatie), *route* (wie (bv verpleegkundige of arts) of wat (bijvoorbeeld folder) gaat met ouders communiceren), *en design* (communicatie stijl; bv objectief en direct of empathisch, coachend) **(Hoofdstuk 7).**

**Hoofdstuk 9 en Hoofdstuk 10** beschrijft een onderzoek naar het effect van een neonatologie afdeling waar geen scheiding tussen ouders en kind plaatsvindt. De focus ligt daarbij op de (mentale) uitkomsten in ouders (vaders en moeders) en hun actieve participatie ten tijde van opname van het kind. Het beschrijft de uitkomsten in ouders die zorg ontvingen met hun kind in het Anna Paviljoen in OLVG (locatie Oost), Amsterdam. In dit zorgmodel, wordt er geïntegreerde zorg in verlos-, kraam- en neonatologie afdeling met familiekamers geleverd, met daarbij volledige couplet-care (moeder-kind zorg) met daarnaast ook het ondersteunen van ouders en hun actieve participatie conform het familie-geïntegreerde zorg (FICare) concept. De studies vergeleken prematuur geboren kinderen en hun ouders opgenomen op de FICare afdeling met kinderen die in een standaard setting, met moeder-kind scheiding (moeder opgenomen op de kraamafdeling, kind opgenomen op de neonatologie afdeling) en zonder FICare concepten werden opgenomen in 2 andere ziekenhuizen. In deze prospectieve niet-gerandomiseerde studie (Mei 2017- Januari 2020, AMICA studie) werden 296 moeders en 263 vaders van 358 prematuur geboren kinderen (mediane zwangerschapsduur bij geboorte 33+2 weken) geïncludeerd. **Hoofdstuk 9** laat zien dat moeders van prematuur geboren kinderen in de innovatieve FICare afdeling minder stress ondervonden en specifiek minder stress door minder moeder-kind scheiding. Actieve participatie in de neonatale zorg was deels een mediator van dit positieve effect van de FICare setting op stress, en specifiek participatie in dagelijkse zorg, medische zorg, tijd doorgebracht met het kind en nabijheid en comfort bieden aan het kind. Participatie, en niet zo zeer de setting met familie-kamers, was een grote mediator voor het gunstige effect van de FICare setting op moeders van prematuur geboren kinderen en voor hen minder depressie en angst, meer zelfredzaamheid en hogere waarden in moeder-kind binding. **Hoofdstuk 10** beschrijft een analyse van de uitkomsten van de vaders van prematuur geboren kinderen. Ook vaders ondervonden minders stress en hadden hogere participatie scores in de FICare setting vergeleken met de standaard zorg afdelingen. Participatie, maar niet de setting met familie-kamers was met name de verklaring voor het gunstige effect van FICare op gunstige uitkomsten met betrekking tot vaders depressie scores en binding met zijn kind. Het concept waarin ouders en kind nooit van elkaar worden gescheiden is daarom dus van belang met betrekking tot de (mentale) gezondheidsuitkomsten en actieve participatie van ouders en hun prematuur geboren kinderen.

Het laatste hoofdstuk (**Hoofdstuk 11**) vat de belangrijkste resultaten samen van de studies die zijn uitgevoerd voor dit proefschrift en bediscussiëert wat dit betekent in het licht van andere studies en de neonatale zorg in zijn algemeen en de relatie tussen ouder-kind nabijheid en “parent-partnered” neonatale zorg modellen. Bovendien bediscussiëert het welke ondersteuning en barrières zorgprofessionals ondervinden om ouders nabij te laten zijn ten tijde van de opname van hun kind op een neonatologie afdeling. Daarnaast bespreekt het welke rol ouder-professional communicatie heeft binnen de neonatologie context. Hierin wordt ook een voorstel voorwaarts ten aanzien van “zero-separation” in de neonatale zorg en hoe professionals en ouders echt met elkaar kunnen samenwerken gedaan, ondersteund met het onderzoek in ons innovatieve FICare model. Daarnaast geeft het uiteindelijk een perspectief ten aanzien van de toekomst en verbetering van de neonatale zorg en onderzoek. De focus blijft daarin de gezondheidsvoordelen die zowel ouders als prematuur geboren kinderen ervaren, wanneer ouders als een volwaardig en uniek teamlid worden geïntegreerd in de neonatale zorg en we ouder-kind scheiding proberen te voorkomen en minimaliseren.

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Publications
List of authors and collaborators
Patient participation in this thesis
Portfolio
Acknowledgments (dankwoord)
Biography

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**Non-author contributions:**

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*School of Nursing*
Prof. dr. L.S. (Linda) Franck

# PATIENT PARTICIPATION IN THIS THESIS

(Veteran) parents had an invaluable role within some of the chapters of this thesis and I learned so much from them. However, I do need to aknowledge the imperfect level of partnership as the partnership was not always present in some of the chapters. This invaluable partnership with parents became clearer to me during the conduct of the research for this thesis. Nowadays, any study or quality improvement intervention that I (would) start or conduct always includes parents from the start.

**Chapter 1, 2, 3, and 4:** no official parent participation.

**Chapter 5:** At the start of the study we included the Dutch veteran parent association of NICU babies (Care4Neo) in development of the interview guide. Veteran parents responded to an online survey on the website of Care4Neo and rated interview questions as: "essential, this really should be asked" "potentially essential, you could consider asking this" and "not essential, don't ask this". Also, parents could add information or questions they deemed essential to be asked in the interviews, which were not present in the questionnaire. In total 48 parents (46 mothers and 2 fathers) participated. Most important issues deemed essential to ask were: if parents get separated from their child (97%), if possibilities for rooming-in were present, what parents could do in care of their infant, if family centred rounds were implemented and if parents could perform SSC (all > 80%). (Potentially) essential questions were subsequently used for the semi-structured interviews. Additionally we performed our study with 2 parent representatives (NHML and SM). NHML is a health communication researcher, specialized in provider-patient relationships and experienced in qualitative research. She is also a parent of a preterm infant, born at 26 weeks of gestation. She experienced mother-infant separation postnatally during her stay in a level 3 NICU, and complete mother-infant closeness (zero-separation) in a level 2 NICU. SM experienced the NICU with very preterm birth, and is co-founder and currently the chairwoman of the executive board of the European Foundation for Care of Newborn Infants (EFCNI). Data analysis was performed by NRvV and NHML and reviewed with a parent representative (SM). Writing and reviewing of the manuscript was done in full collaboration and partnership with SM and NHML.

**Chapter 6, 9 and 10:** We included parents in the item generation phase of the CO-PARTNER tool using purposive sampling in May 2016-April 2017. Here I acknowledge, that parents were more used as a research subject, but along the way they became more of an advisor to the conduct of my study on developing the CO-PARTNER tool and the AMICA-study, than solely a research subject. However, this is not formally captured within the studies as written in this thesis. During screening for inclusion and informal conversations with (veteran) parents their view became more and more important, and even though the study was already set up and ready to run in April 2017 their informal feedback and suggestions during conduct of the AMICA study was invaluable. Before conduct of the AMICA study, we initialized focus groups, one-on-one interviews and scoring of the initial instrument for parent participation with a purposive sample of forty-five parents. Parents (mothers or fathers >18 years of age) had a preterm infant (born at a gestational age between 24 weeks - 36 6/7 weeks), were at the time experiencing or had experienced a NICU stay in the previous 2 years, and had experience in either a standard or FICare unit participating in the AMICA trial. Participants were asked to identify (additional) items on parent participation. Above, we investigated their views on content of items, how response options to items should be presented and on the rightful inclusion of the 26 items from the original Index of Parent Participation in the first version of the tool. Participants were asked to score items (during generation from the original Index of Parent Participation, focus groups or one-on-one interviews) as; (1) relevant or not relevant in light of parent participation in the NICU; (2) if the items needed a yes/no response, or if the items had to be scored on a scale and were intended to examine a collaborative process in care towards being able to perform activities independently ('the nurse does this', 'the nurse and I do this together' and 'I do this independently'). After the AMICA study started, the parents too were included to study the effect of an innovative FICare model on their mental health.

**Chapter 7 and 8**: NL assumed a dual role of investigator-parent representative from inception to completion of these studies, being a mother to a daughter who was born at 26 weeks' gestation. Two parent representatives provided additional feedback on our analyses and manuscripts. These are the founders of a Dutch parent-support platform (Kleine Kanjers) and a parent-support organization for parents of preterm infants (Veerkrachtige Ouders).

# PORTFOLIO

| | Year | Workload (ECTS) |
|---|---|---|
| **General courses** | | |
| Good Clinical Practice (OLVG) | 2016 | 1 |
| Searching for evidence (AMC) | 2016 | 0.2 |
| Presenting techniques (AMC) | 2016 | 0.2 |
| Clinical Data management (AMC) | 2016 | 0.2 |
| Project management (AMC) | 2017 | 0.2 |
| Scientific writing in English (AMC) | 2018 | 0.2 |
| Basiscursus Regelgeving en Organisatie voor Klinisch Onderzoekers (eBROK) | 2019 | 1.5 |
| **Specific courses** | | |
| Practical Biostatistics (AMC) | 2016 | 0.5 |
| Master in Epidemiology and Biostatistics (EpidM, VU University), (MSc, Cum laude)<br>Specific courses in:<br>Longitudinal data-analysis<br>Missing data analysis<br>Mediation analysis<br>Systematic Reviews and Meta-analyses | 2016 - 2019 | 60 |
| Advanced Biostatistics (AMC) | 2018 | 0.5 |
| Introduction into R (OLVG) | 2018 | 0.5 |
| **Seminars, workshops and master classes** | | |
| Panel member on symposium on Family Integrated Care (Nieuwegein, The Netherlands) | 2016 | 0.2 |
| FAMElab: introduction in pitching of research (AMC) | 2017 | 0.2 |
| Organization Family Integrated Care Symposium (OLVG) | 2019 | 0.5 |
| External expert in Core Outcome Sets for Infants with Fetal Growth Restriction (Brighton, UK and Maastricht, Netherlands) | 2018 – 2019 | 0.5 |
| Developing and Validating Metrics and Measures for Stakeholder Engagement in Research (National Institutes of Health Webinar) | 2020 | 0.1 |
| **Presentations at (inter)national conferences** | | |
| Wetenschapsdag OLVG (poster) | 2016 | 0.5 |
| Nutrition and Growth (oral, Amsterdam, the Netherlands) | 2017 | 1.5 |
| Amsterdam Kindersymposium | 2017 | 0.25 |
| Amsterdam Kindersymposium | 2018 | 0.25 |
| World Association on Infant Mental Health (2 oral, 1 poster, Rome, Italy) | 2018 | 1.5 |

| | Year | Workload (ECTS) |
|---|---|---|
| Separation and Closeness Experiences in the Neonatale Environment (SCENE, Lecco, Italy) | 2018 | 0.5 |
| Invited speaker, National Nursing conference on caring for infants at risk (oral, Ede, the Netherlands) | 2019 | 0.5 |
| Nutrition and Growth (1 oral, Valencia, Spain) | 2019 | 0.5 |
| Pediatric Academic Societies (2 posters, PAS, Baltimore, USA) | 2019 | 1 |
| joint European Neonatal Societies (3 oral, Maastricht, Netherlands) | 2019 | 1.5 |
| Invited speaker, International Conference on Family Integrated Care (oral, Leeds, UK) | 2019 | 1 |
| SCENE (1 oral, 2 posters, Budapest, Hungary) | 2019 | 1.5 |
| Amsterdam Kindersymposium | 2020 | 0.25 |
| COSGROVE jENS and Brighton invited expert | 2018 – 2020 | 0.5 |
| British Association of Perinatal Medicine – BAPM webinar - Getting to Zero Separation: Prioritising Family Integrated Care for our new normal (Invited speaker) | 2020 | 0.5 |
| CPBF WEBINAR PREEMIE POWER WEEK (Invited speaker) | 2020 | 0.5 |
| KINDERfonds Keeping Families Close (Invited speaker) | 2020 | 0.5 |
| Vakblad Vroeg Wereld Prematurendag (Invited speaker) | 2021 | 0.5 |
| Expert committee core outcome set generation for family centered and integrated care | 2021 – present | 0.5 |
| European Association of Pediatric Societies, Barcelona (Invited speaker) | 2022 | 0.5 |
| Invited speaker at the opening of the new NICU of University of Ghent | 2022 | 0.5 |
| World Prematurity Day – NICU partners in Care (Baxter, Invited speaker) | 2022 | 0.5 |
| International Family Integrated Care Conference (Invited speaker, Session Chair) | 2022 | 0.5 |
| TULIPS Jonge OnderzoekersDag – Research in non-academic centers (Invited speaker) | 2022 | 0.25 |
| **Other** | | |
| Student coaching / mentoring: 8 Bachelor of Science students (3 months) | 2016 – 2020 | 8 |
| Student coaching / mentoring: 20 Master of Science students (4 months) | 2016 – 2020 | 27 |
| Member on national guideline committee for Postnatal care (NVK) | 2016 – 2020 | 4 |

| | Year | Workload (ECTS) |
|---|---|---|
| Founder and board member of Researchers of OLVG (OvO) | 2017 – 2020 | 3 |
| Organiser, supporter and presenter of recurring epidemiologic masterclasses in OLVG | 2017 - 2020 | 3 |
| Poster Jury for Wetenschapsdag (OLVG) | 2017 | 1 |
| Organisation of Wetenschapsdag (OLVG) | 2018 | 1 |
| Peer Support (OLVG) | 2018 | 0.5 |
| Editorial board member of Wetenschap@OLVG | 2018 – 2021 | 3 |
| International Steering Committee on Family Integrated Care | 2019 – present | 4 |
| Supporting PhD and resident colleagues with epidemiologic questions/research | 2019-present | 1 |
| Training Upcoming Leaders in Pediatric Sciences (TULIPS, NVK, class 2019-2021) | 2019 – 2021 | 4 |
| Marie Sklowdowska-Curie Action for R&I Staff Exchange (RISE) scheme: FAMinCare: Integrating families at NICUs to empower them as primary caregivers to improve the health outcomes of their high-risk neonates (grant appointed) | 2019 – present | 1 |
| Conceptualisation and writing of grant application: Parents as Partner in Neonatal care, a stepped-wedge cluster randomised trial: the neo-PARTNER study (grant appointed) | 2019 – present | 2 |
| Organiser, supporter and presenter of Journal Club/Watch AmsterdamUMC | 2021-present | 1 |
| **Parameters of Esteem** | | |
| Innovation prize OLVG | 2018 | |
| Best clinical research project of OLVG | 2019 | |
| Prijs voor de Jonge Onderzoeker, Nederlandse Vereniging voor Kindergeneeskunde | 2020 | |
| Nominee for the Young Investigator Award (European Society for Pediatric Research) | 2021 | |
| **Total** | | **146** |

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# BIOGRAPHY

Nicole was born on October 30th 1988 in Aalsmeer, the Netherlands to Joost and Teri van Veenendaal. From a very young age she enjoyed and developed an interest for research. At the age of 8 she owned a microscope and would determine butterflies, different plants in the garden, and collect tadpoles to nurture them (not always successfully) to fullblown toads, with the intention to become a biologist. The family house was full of experiments and scientific games, including examining human behavior towards changes in usual appearance of food, experimenting with chrystals and memorizing all capitals in the world. During elementary school Nicole would write (extra) essays on the ancient Egyptians and deep sea life. In high-school she would study the origins of allergies and for the first time get into contact with an academic medical doctor, who would inspire her and give her perspective for the future: to become a doctor and researcher.

She completed her Gymnasium studies in 2007 (*cum laude*) at the Alkwin Kollege, Uithoorn, the Netherlands, and set off to study Medicine at the University of Amsterdam in the Netherlands. She enrolled into the Honoursprogramme (supervised by prof. dr. C.M. (Carla) Hollak and prof. dr. C. (Carel) Noort). Her first experiences with research were with dr. K. (Kees) Boer at the department of Gynaecology and Obstetrics, on the origins of hypertryclyceridemia in pregnant women on highly-active antiretroviral therapy for HIV. After, she studied the natural history of patients with Morquio type IV disease (department of Pediatric Metabolics), and got into contact with her first laboratory experiments, trying to grow cartilage *in vitro* (technically very challenging).

Next, she set off to the University of Yale (New Haven, CT, USA) in 2010, with a Huygens Talent Scholarship (provided by the Ministry of Education, Nuffic). There she studied the association between treatment of pediatric Graves' disease in children and the development of obesity, and the teratogenic potential of antithyroid drugs (under supervision of prof. dr. S.A. (Scott) Rivkees), examining tadpoals in the laboratory. For this she was nominated the University thesis prize in 2012 and won the Student Research Award at the Pediatric Academic Societies (PAS, Boston, USA). She continued her studies within the department of Pediatric Genetics (University of Groningen and University of Amsterdam) studying the association of antithyroid drugs with heterotaxy syndrome in pediatric patients (under supervision of dr. J.M. (Jan-Maarten) Cobben and dr. J.E.H. (Jorieke) Bergman-van Kammen)).

In 2014 she completed her studies in Medicine (MSc, *cum laude*), and Pediatrics and Neonatology sparked her interest. She started her residency (ANIOS) period in OLVG, a large teaching hospital in Amsterdam, the Netherlands (under supervision of dr. A.A.M.W. (Anne) van Kempen and drs. H.C. (Carlijn) Kraakman). In 2016 she set up a research group together with Anne and dr. S.R.D. (Sophie) van der Schoor (under supervision of prof. dr. J.B. (Hans) van Goudoever) on Family Integrated Care. She joined the working group for the National Guideline on Postnatal Care for Infants (under supervision of dr. L.M. van der Meer). She completed a degree in Epidemiology and Biostatistics during her PhD-studies (under supervision of prof. dr. J.W.R. Twisk, MSc, *cum laude*) and became a participant in the Training Upcoming Leaders in Pediatric Science (TULIPS) program. During her studies she worked as a doctor in the very busy pediatric and neonatal clinic of OLVG, keeping up her clinical, social and practical skills, and in 2021 she started her training to become a pediatrician in the Amsterdam University Medical Centres (under supervision of dr. D.K. Bosman and dr. H.M.A. de Bie). She currently lives with Haico and their daughter Lena in Almere (near the city of Amsterdam) where she can be regularly found sailing the lakes surrounding the city.