Effect of Family-centered Care Interventions on Motor and Neurobehavior Development of Very Preterm Infants: A Protocol of Systematic Review

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Protocol

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Abstract

**Background:** Globally, preterm birth is a health concern leading to various developmental difficulties such as poor motor and/or cognitive function. For infants born preterm, FCC promotes developmental skills over the time in an appropriate enriched environment. The purpose of this study is to systematically review and assess the evidence of FCC interventions on motor and neurobehavioral development in very preterm infants. Additionally, this review aims to determine the factors that might affect the infant development.

**Methods:** Systematic review will be carried out by including quasi-experimental controlled trials and randomized controlled trials. Electronic databases such as Scopus, PubMed, EMBASE, Cochrane Library, Web of Science, CINAHL, and PsycINFO will be searched using database specific terms. Additionally, searches will be carried out in ProQuest, and references of included studies will be searched. Two review authors, independently, will conduct the screening, data extraction, and critical appraisal of included studies. If possible, meta-analysis will be undertaken to assess the effect of FCC on motor and neurobehavior of premature infants.

**Conclusion:** The review will provide insights regarding the effect of the FCC on preterm infants. This systematic review will guide the clinicians on the feasibility of practicing FCC that might support and promote the integration of parents into various rehabilitation settings.

**Systematic review registration:** Protocol has been submitted to PROSPERO on July 26, 2020.

**Background**

**Description of the condition**

Preterm birth is described as any birth ≤37 weeks of gestation [1]. Globally, approximately five to 18% (15 million) of neonates are born preterm of all the live births [2]. Very preterm babies born less than or equivalent to 32 weeks of gestation are at high-risk of impaired cognitive, sensory, motor, emotional, language, and behavioral development when compared to the full-term infants [3]. Early brain injury and impaired brain development are reasons for the aforementioned problems [4].

In the first year of life, biological, environmental, and social factors influence the development of early motor and neurobehavior development [5]. Motor development encompasses quality of movement, developmental milestones, motor skills, visual-spatial, visual-motor integration, balance, and coordination [5]. Neurobehavioral development occurs with efficient responses to the sensory (tactile, auditory, visual, olfactory) stimulation and autonomic nervous system, organization of state (calm, excited, irritable), and self-regulation (hand to mouth responses); language, attention, socio-emotional development and executive function [6].
Very preterm infants are usually admitted in the neonatal intensive care units (NICU) to provide special care and management. In the NICU, very preterm infants face many stressful situations such as excessive sound, bright light, painful medical applications, and lack of parental contact, which they would not have experienced in utero [7]. Excessive sensory load on tactile, olfactory, gustatory, visual, and auditory systems during this critical period of brain development impairs the baby’s physiological responses and may impact negatively on their neuromotor and behavioral development [8]. Therefore, the relationship between preterm birth and the aforementioned chain of events might affect the development of motor skills and neurobehavior in the later span of life [7].

Family and home-based environments are considered to have the greatest and permanent effect on an infant’s growth within the ecological system, even when taking into consideration of certain other aspects such as education and socioeconomic status of the parents [9]. The birth of a very preterm infant affects the mental well-being of the parents, leading to distress and anxiety and alters the sense of bonding and parenting skills to care [10]. A review on the effects of early intervention for very preterm infants found that early intervention strategies addressing the relationship between parent-child was effective when compared to interventions centered on the child or the parent alone. It is necessary to educate and involve the family in the treatment of infants to have a greater effect on their developmental outcomes [11]. Therefore, strategies focused primarily on responsive parenting might promote the growth of the child through family dyadic relationships [12].

**Description of the Intervention**

Family-centered care (FCC) is described as a healthcare approach that involves planning, delivery, and evaluation centered on equally positive relationships among families, healthcare providers, and patients [13]. FCC’s values include family care, inclusive family engagement, interaction, empathy for and integrity of families, and knowledge transformation [11]. It has been implemented in developed and a few of the developing countries [14]. FCC can be practiced in various settings such as homes, clinics, hospitals, and communities. The central focus of FCC is to train the parent as a primary therapist and provide psychosocial support. It is a promising approach in newborn care especially for very preterm infants to achieve short-term and long-term benefits [15].

**How the intervention might work**

Many FCC approaches are delivered through family or parents and include various components of developmental care, [16] e.g. Newborn Developmental Care and Assessment Program (NIDCAP) [17] and Creating Opportunities for Parent Empowerment (COPE) [18]. Studies have shown the importance of early developmental interventions in improving neurobehavioral outcomes in preterm infants [19]. Interaction between an infant and parent begins before infancy and is greater at childhood, with social-emotional experiences involving gestures, cries, smiles, mutual gaze vocalizations and continues throughout childhood [20]. A significant aspect affecting the growth of the child is the quality of interaction between an infant and parents. Modification of an infant’s physical and emotional environment both in the NICU and post-hospitalization at home will have positive outcomes in their overall development [21]. The FCC
has shown an increased knowledge, capacity, and competence of the parents to care for their infant or child [22]. The potential benefit of the FCC is that it is relatively low cost and has the focal involvement of parents for a long-term period. It has shown to decrease the length of hospitalization, improve the wellbeing of preterm by allowing better allocation of resources, and enhancing parent-infant bonding [23].

Why it is important to do this review

In infants born very preterm certain aspects of motor and neurobehavior function are frequently impaired in relative to their counterparts born at term. FCC has been found to support the infant’s care by enriching the environment, improving cognitive and physical growth and further promoting early developmental resilience (right from the birth) [24, 25]. One of the review described that family-based interventions reduces preterm parental stress and it has cost-effective benefits when practiced in the NICUs [24]. Another review reported, FCC interventions for preterm infants in NICU have shown to improve their weight gain, reduce hospital readmission, and provide positive parental outcomes (such as parent satisfaction, reduced anxiety, and stress) [12]. Literature suggests that a supportive family environment, a positive bond among parent and infant leads to improved neurodevelopmental outcomes even if the preterm infants are exposed to vulnerabilities such as neurological abnormality [26, 27]. The studies have shown that negative and intrusive parenting leads to poor developmental outcomes in preterm children across childhood. On the contrary, warm, sensitive, and positive parenting might result in a protective effect on the preterm infant’s development [28].

However, to the best of our knowledge, no systematic review has studied the impact of FCC interventions on the motor and neurobehavioral function in very preterm infants during both the NICU stay and the follow-up period. Furthermore, the various factors (mode of delivering the intervention, dosage, etc.) that might influence the success of FCC are not yet known. Hence, we would anticipate that the findings from this review will help inform clinicians, parents, and educators about the role of the FCC in promoting motor and neurobehavioral development in preterm infants. With this background, we have proposed two research questions; 1) What is the evidence of data available on the impact of FCC on very preterm infants when compared to standard care/ interventions without involving family on motor and neurobehavior development? 2) What are the factors that determine the infant development due to FCC interventions?

We have described the relationship between the very preterm infant and FCC along with their possible outcomes through conceptual framework (Fig. 1). In NICU, if there is limited parent-infant interaction, this might result in parental stress and poor bonding between them. This might further impact on delay or poor motor and neurobehavioral development of the infants and increases the level of parent anxiety and stress. On the other hand, involving parents or implementing the FCC right from the beginning in the NICU might create a well nurturing environment and a positive parent-infant bonding due to the effect of various sensory experiences and activity-dependent brain activation. Further, this might accelerate the development of motor and neurobehavioral function along with the increased ability of learning and memory in preterm infants.
Methods

Review registration and reporting

The systematic review is submitted to PROSPERO (the International Prospective Register of Systematic Reviews) for registration on July 26, 2020. We have adhered to PRISMA-P (Preferred reporting items for systematic review and meta-analysis) guidelines to report this protocol (supplementary data file 1).

Eligibility criteria

Types of studies: Quasi-experimental trials and randomized controlled trials are eligible to be included. Non-randomized trials, observational studies, study protocols, editorials, reviews, conference abstracts, letters, and commentaries will be excluded.

Type of participants: Preterm infants born lesser than 32 weeks of gestation and their primary caregivers. Primary caregivers can be either mother, father, or grandparents.

Place of recruitment: This review does not impose any restrictions on the setting. It can be in the NICU, hospital, and home or community.

Time of recruitment and follow up: The studies should have recruited infants from birth in the NICU stay to six months, and followed up till two years of age.

Intervention: This review will include any FCC interventions involving the establishment of a collaborative relationship between the healthcare professional and the parent, mutually agreed-upon goal setting, creating the home program by selecting therapeutic activities that focus on accomplishing family objectives, supporting the implementation of the program through home visits, parent education and evaluating the outcomes. Interventions having at least two components of FCC will be included. The intervention will involve supervision and support from a clinician or professional such as a neonatologist, pediatrician, nurse, physiotherapist, occupational therapist, speech-language pathologists, and other rehabilitation team members.

Type of comparators: The studies should have compared the FCC to the therapist provided standard care interventions or usual care.

Type of exposure: such as age, time of recruitment and follow up, settings, FCC providers, intensity and frequency of intervention; parental behavior, responsivity, and parental satisfaction that may influence infant development.

Types of outcome measures: Primary outcomes - We will include studies that measure motor and neurobehavioral function.

Motor functions such as quality of movement, gross and fine motor skills, developmental milestones, visual-spatial, visual-motor integration, balance, and coordination are measured through any of the
outcome measures. The outcome measures could be Prechtl's General Movements Assessment (GMA), Test of Infant Motor Performance (TIMP), Alberta Infant Motor Scale (AIMS), Neuromotor Behavioral Assessment (NMBA), Hammersmith Infant Neurological Examination (HINE), Pediatric Evaluation of Disability Inventory (PEDI), Peabody Developmental Motor Scale (PDMS) and Bayley Scale of Infant and Toddler Development (BSID).

Neurobehavior is measured in terms of the sensory and autonomic nervous system, organization of state (calm, excited, irritable) and self-regulation (hand to mouth responses); language, attention, socio-emotional development, and executive function using any of the outcome measures. Neurobehavior could have measured using Assessment of Preterm Infants Behavior (APIB), Brazelton Neonatal Behavioral Assessment Scale (NBAS), Neurobehavioral Assessment of Preterm Infants (NAPI) and NICU Network Neurobehavioral Scale (NNNS).

Secondary outcomes are changes in parental behaviors or responsivity captured through videotaped interactions or observations and measured by any of the validated scales. Parental satisfaction will be measured by questionnaires and interviews. Factors such as age, time of recruitment and follow up, settings, FCC providers, intensity, and frequency of intervention which might influence infants development will be considered for the review.

**Search methods for identification of studies**

Electronic databases: We will search PubMed, Cochrane Central, Scopus, EMBASE, PsycINFO (Ovid SP), CINAHL and Web of Science. Articles that are written in English from January 2010 to August 15, 2020 will be included. The following keywords will be used; “family-centered care”, “family-centric approach”, “preterm infants”, “motor development”, “neurobehavior development”.

Searching other resources: We will search in ProQuest, ClinicalTrials.gov, EU Clinical Trials Register, metaRegister of Controlled Trials, and the World Health Organization (WHO) International Clinical Trials Registry Platform search portal. We will contact field experts and corresponding researchers of included studies to get relevant additional information. To find related studies, we shall search for reference lists and forward citations of included studies. Identified records will be exported to EndNote X7 for data management.

**Selection of studies**

Two reviewers will search and read the titles, abstracts of the listed sources individually, and exclude any studies depending on the eligibility criteria. If disparity arises between the authors, it will be resolved by discussions and will be reviewed as to their full text. After obtaining the full text of the included abstracts, three review authors will independently rank these as ‘include’ or ‘exclude’. If necessary, we will address any differences in agreement, with a senior review author. We will record the reasons for excluding the articles. Proposed screening protocol for abstracts and full texts has been attached as supplementary data file 2. Reference details, any knowledge accessible on current research and record details on similar
publications such that each study will be considered as the unit of concern in the analysis. An adapted PRISMA will be implemented (Fig. 2).

Data extraction and management

The extraction of general characteristics such as study identifiers, location of the study, participants, study selection criteria, and outcomes from the included studies will be carried out. The TIDier (Template for Intervention Description and Replication) checklist will be used to summarize the list of intervention characteristics and assist in the replicability of interventions and comparability between the studies [28]. Independently, two review authors will abstract the data from the articles included in the review using the data extraction templates. From each of the included studies, the following information will be extracted: study identification (title and authors), the country of study, year of publication, sample size, features of intervention, outcomes, evaluation tools, results, and conclusions. In case of disagreement, we will discuss until consensus or with the help of the third reviewer, any disagreements will be resolved.

Addressing redundant and related publications

In the case of redundant papers, related records, or several primary research studies, we can optimize the data yield by gathering all the relevant details by utilizing the highly broad dataset collected through each of the documented papers. In correlation with our results, the publication which reports the longest follow-up will be considered as a priority.

Risk of bias assessment among included studies

The Revised Cochrane Risk of Bias Tool (RoB 2.0) would be employed to critically appraise the quality of the included articles. The RoB 2.0 tool assesses the randomization process, deviations from the intended protocol, measurement of the outcome, missing outcome data, and selective reporting. Two authors will appraise all the articles, independently. There should be agreement about the existence of certain inconsistencies. The risk will be categorized into low risk, high risk and some concern [30].

Addressing the missing data

From the authors missing information would be collected and significant empirical data such as screened, randomized, intention-to-treat, as-treated, and per-protocol population will be closely analyzed. If authors will not respond within 15 days of last communication (email), the study would be excluded from the review. We will critically appraise the concerns related to the missing data and imputation methods, e.g. Last Observation Carried Forward (LOCF). The attrition rates, e.g. drop-outs, losses to follow up and withdrawals will be investigated.

Data synthesis

The outcomes will be presented as the mean differences (MDs) / standardized mean differences (SMDs) with 95% confidence intervals (CI) unless otherwise stated for continuous variable. The odds ratios (ORs) or risk ratio (RRs) with 95% CI will be expressed as the dichotomous data. If there is strong evidence of
homogeneous effects through findings, using a model of random effects we will mainly sum up a low probability of bias results. The meta-analysis of random effects will be viewed with due consideration of the full range of outcomes, preferably by providing an interval of predictions. In each study, the predicted range for the true treatment effect will be specified using the prediction interval. The quality of evidence of the included studies in the review will be assessed using Grading of Recommendations Assessment, Development and Evaluation (GRADE) system. This system grades quality of evidence at 4 levels: high (4), moderate (3), low (2), and very low (1) [31].

**Subgroup analyses and investigation of heterogeneity**

In the meta-analysis, we will not record the findings of the sample as the pooled effect estimate if there is any the event of major scientific, analytical, or empirical variability. Using a standard Chi-square test (significance level of $\alpha = 0.1$) we will recognize the heterogeneity. Using the $I^2$ statistics, inconsistency across studies will be quantified. It is considered to be a high level of inconsistency if an $I^2$ statistic is 75% or more. If there is significant variability, we will aim to identify potential explanations for this by analyzing the features of the specific study and other subgroups. Subgroup analyses of factors such as gender, age, country, type, and mode of intervention and outcome measures will be carried out to explore the interaction between them.

**Sensitivity analysis**

To understand how the following factors influence the effect sizes, we will perform the sensitivity analyses. The factors include, the risk of bias of included studies; large and long trials to understand the extent to which they influence the results. Different effect size measurements such as risk ratio and odds ratio along with various statistical models such as fixed-effect and random-effects models will be used to test the robustness of the results.

**Narrative synthesis**

When it is not possible to conduct the meta-analysis because of significant statistical, clinical or methodological heterogeneity, a narrative synthesis will be done. Studies would be narratively defined focusing on the intervention and outcomes. The subsequent results would be summarized using the tables and figures.

**Discussion**

Through FCC the health professionals guide the programs, work in partnership with the parents to support and guide the infant’s motor and neurobehavior development to enhance their capabilities. Providing care to the preterm infants through a family-centered approach, may improve the overall infant development and in turn reduces the burden of the caregivers and enhances their capacities. Findings of this review will provide understanding effectiveness of FCC components, its benefits on very preterm infants and thereby help policymakers and health professionals to adapt evidence based decision making and practice of FCC. This systematic review will guide the clinicians on the feasibility of
practicing FCC that might support and promote the integration of parents into various rehabilitation settings.

**Strengths And Limitations**

To the best of our knowledge, this systematic review is the first to address the effect of FCC on improving motor and neurobehavioral outcomes in preterm infants and the factors influencing the infant development. We will undertake comprehensive search in various databases to identify the studies however, due to resource constraint studies published in English will be considered.

**Abbreviations**

FCC  
Family-centered care  
NICU  
Neonatal Intensive Care Unit  
PROSPERO  
The International Prospective Register of Systematic Reviews  
NIDCAP  
Newborn Developmental Care and Assessment Program  
COPE  
Creating Opportunities for Parent Empowerment  
PRISMA-P  
Preferred Reporting Items for Systematic Review and Meta-analysis  
GMA  
General Movements Assessment  
TIMP  
Test of Infant Motor Performance  
AIMS  
Alberta Infant Motor Scale  
NMBA  
Neuromotor Behavioral Assessment  
HINE  
Hammersmith Infant Neurological Examination  
PEDI  
Pediatric Evaluation of Disability Inventory  
PDMS  
Peabody Developmental Motor Scale  
BSID  
Bayley Scale of Infant and Toddler Development
APIB
Assessment of Preterm Infants Behavior

NBAS
Neonatal Behavioral Assessment Scale

NAPI
Neurobehavioral Assessment of Preterm Infants

NNNS
NICU Network Neurobehavioral Scale

TIDier checklist
Template for Intervention Description and Replication

ROB 2.0 Tool
The Revised Cochrane Risk of Bias

LOCF
Last Observation Carried Forward

WHO
World Health Organization

MD
Mean difference

SMD
Standardized mean difference

OR
Odds ratio

RR
Risk ratio

CI
Confidence Interval

GRADE
Grading of Recommendations Assessment, Development and Evaluation

Declarations

- **Ethics approval and consent to participate**: Ethical approval is not applicable as this is a systematic review protocol, we will not involve human population directly.

- **Consent for publication**: Not applicable as the manuscript does not contain data from individual person.

- **Availability of data and materials**: The data acquisition for this systematic review has not yet started. We plan to conduct the search in mid-August. Subsequently, dataset generated through this systematic review can be requested from corresponding author.

- **Competing interests**: The authors declare that they have no competing interests.
• **Funding:** Authors declare no source of funding.

• **Author contributions:** BK and MK conceiving the topic. SSP, BK, AS and MK designed the protocol. BK coordinated with the team and timeline to finalize the protocol. BK, SR, MK, AS and SSP designed the search strategies. MK, BK, and SSP wrote the protocol. All authors read, provided general advice and approved the final version of the protocol. BK is the guarantor of the review.

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**References**


Figures
Figure 1

Conceptual Framework of family-centered care on motor and neurobehavior development of preterm infants
Figure 2
Flow chart of study selection

Supplementary Files

This is a list of supplementary files associated with this preprint. Click to download.

- ScreeningprotocolforAbstractsFulltexts.docx
- PRISMAPchecklist.docx