Advancing early intervention for preterm infants

The innovation-impact cycle of the TOP program

Monique Flierman

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As you start to walk on the way, the way appears.

Rumi/Masnavi,
adapted by C. Barks

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Chapter 1

Introduction and outline of the thesis

Prematurity continues to pose substantial challenges for early childhood development, as children born preterm are at increased risk for cognitive, motor, behavioral, and socio-emotional difficulties. Over the last decade, the TOP program provides home-based responsive parenting support and has reached thousands of families since its nationwide implementation. As neonatal care and family needs evolve, early-intervention programs must be continuously refined to remain effective, accessible, and scalable. This thesis contributes to that ongoing innovation by enhancing execution quality, broadening the program's reach, and integrating new digital tools to strengthen support for preterm infants and their families.

Prevalence of preterm birth

Preterm birth (PT), defined as delivery before 37 weeks of gestational age (GA), remains a significant global health challenge. Each year, an estimated 13.4 million babies are born preterm, representing 9.9% of all live births worldwide (1-3). In the Netherlands this percentage is lower: 6.3% of all live born infants are born preterm, with the following distribution: very preterm (VP); 28–32 weeks GA: 0.8%; and moderate to late preterm infants (MLP), typically defined as those born between 32 and < 37 weeks of GA, representing a significant proportion of all preterm births: 5.5% (4).

Short- and long-term consequences of preterm birth

Preterm birth is a significant cause of neonatal morbidity and mortality worldwide. Advances in neonatal care have significantly improved survival rates among PT infants. However, PT children remain highly vulnerable to a wide range of short-and long-term adverse health, motor, cognitive, and behavioral outcomes (5, 6). The prevalence and severity of complications and co-morbidities correlate closely with the degree of prematurity, increasing as GA decreases (7-13).

Neonatal complications

In the health domain, VP infants often face significant neonatal complications including bronchopulmonary dysplasia (BPD), intraventricular hemorrhage, and neonatal sepsis (14). These early medical challenges are not only predictive of increased morbidity but also of later neurodevelopmental impairments (5).

Although most research focuses on VP infants compared with term infants, MLP infants are also at significant risk for increased health problems during and after the hospital period, including hypothermia, hypoglycemia, hyperbilirubinemia, respiratory distress, and poor feeding. Which translate into markedly higher rates of early rehospitalization for jaundice and feeding problems compared to term infants (15). Beyond these acute complications, MLP infants use significantly more health care services across hospital, community, and outpatient settings in the first years of life, generating societal costs up to three to six times higher than costs for term infants (16, 17). Beyond the economic burden, the high number of health care appointments can create a significant burden for parents. Together, these findings highlight a clear need for early-intervention support following discharge to stabilize infant health, reduce preventable readmissions, and protect emerging parent–infant relationships.

Self-regulation

Behavioral and emotional self-regulation defined as the ability to modulate own reactivity and control behavior appropriately is another area of concern for preterm infants. This includes difficulties modulating arousal states, limited capacity for self-regulation (18, 19). This can lead to inconsolable crying, irregular sleep-wake patterns, frequent nighttime awakenings, and poor adaptability to environmental changes (20-22). In addition, PT infants more often exhibit sensory processing disorders, manifesting as hyperresponsivity to auditory, tactile, and visual stimuli, which may contribute to irritability and overstimulation during daily caregiving interactions. Delays or difficulties in the maturation of these regulatory capacities have important implications for later attention regulation and social reciprocity during caregiving and play (23).

Motor development

Motor development is also markedly affected by preterm birth (24). In general, PT infants exhibit more delayed postural control and motor development, with slower achievement of key milestones such as rolling, crawling, and walking (25). However, the developmental trajectories studied by Suir et al. (26, 27) suggest that not only the timing but also the pace and stability of motor skill acquisition in the first year are critical for understanding long-term motor outcomes and planning early support. Meta-analytic findings confirm lower standardized motor scores persisting through childhood, with more pronounced delays in children born at lower gestational ages (28-30).

Neurobehavioral disorders

Prematurity has enduring neurobehavioral consequences that extend well beyond infancy. The prevalence of neurobehavioral disorders such as attention-deficit/hyperactivity disorder (ADHD), autism spectrum disorder (ASD), and internalizing mood disorders is markedly higher among those born preterm (9, 31-33). These neurodevelopmental vulnerabilities are compounded by social difficulties frequently reported in adolescence, including social withdrawal and fewer close friendships (5), and continue into adulthood, reflected in lower educational attainment, reduced employment rates, and decreased likelihood of forming romantic partnerships or becoming parents (34-40).

Cognitive development

Cognitive development is consistently and significantly affected in preterm populations. Meta-analyses among VP infants show lower full-scale IQ, performance IQ, and verbal IQ, with a 10 to 13 points difference compared to their term-born peers (41, 42) and are nearly three times more likely to require special education services (43). Although the differences in performances between MP born and FT born children are smaller, MP children are disadvantaged compared with their full-term classmates (44).

Impact on parenthood

Giving birth to a preterm born infant places immense stress on parents, who must cope with the emotional impact of preterm birth, and at the same time need to develop confidence in their caregiving abilities and parenting skills in a hospital setting (45, 46). The emotional challenging preterm birth is associated with higher levels of stress, anxiety, depressive and post-traumatic stress symptoms, particularly in the early postpartum period (47, 48). The psychological challenges observed during the neonatal period often extend beyond hospital discharge; parents frequently continue to experience elevated distress, and adjustment demands in the home setting (49-51). These difficulties can amplify by the more diffuse behavioral cues, fragmented regulation, and heightened soothing needs often seen in preterm infants, which place greater demands on parents' co-regulation skills (19). Responsive, attuned caregiver–infant interactions can help buffer these risks during a period of heightened developmental sensitivity (52-54).

Parental self-efficacy (PSE), grounded in Bandura's social cognitive theory (55), has been identified as a key determinant of parental and child well-being. PSE refers to parents' beliefs in their ability to influence their child's development in a positive and health-promoting manner. Stronger parental self-efficacy is associated with positive maternal psychological well-being, reduced family stress, and improved responsiveness and parenting behavior. Lower parental self-efficacy has been linked to greater difficulties in coping and reduced responsiveness to infant cues, thereby posing risks for the parent–infant relationship (55-59). According to Bandura's framework, self-efficacy beliefs shape by doing something and succeeding at it, observing others, encouragement from others and paying attention to physical reactions and emotions when facing challenges. Interventions that strengthen these sources of efficacy help to develop a stronger sense of competence, which in turn benefits both parental mental health and child development.

Although Bandura's foundational work predates the emergence of mHealth, digital interventions can also reinforce efficacy. Garfield et al. demonstrated that parents provided with a mobile health application (NICU2HOME) reported consistently higher levels of self-efficacy from admission through 30 days post-discharge compared with controls, with greater app use linked to stronger outcomes (60, 61).

Post-discharge care and the role of parental interventions

The transition from the Neonatal Intensive Care Unit (NICU) to home represents a profoundly vulnerable period for families of preterm infants. Parents often encounter increased caregiving insecurities and significant gaps in knowledge regarding the behavioral and developmental needs of their child. Growing evidence stresses that providing early, structured, and individualized support during this critical phase is essential to promote optimal developmental outcomes and parental well-being (62-64). The most recent Cochrane systematic review by Orton et al. (7) offers high-quality evidence supporting early post-discharge developmental interventions. Systematic reviews and international guidelines underscore that early parental responsiveness is a crucial determinant of child development, and that interventions enhancing parental knowledge, sensitivity, confidence, and emotional regulation have a significant positive impact on PT child outcomes. Early interventions that incorporate both infant-focused strategies and enhancements of the parent-infant relationship are considered most

effective. This dual-focus approach likely exerts its effect through interrelated mechanisms (65). First, enhancing parental sensitivity to infant cues, promoting timely and appropriate responses, supports cognitive and socio-emotional development. Second, improving parental mental health, which is an independent predictor of child development, will also improve neurodevelopmental and behavioral outcomes.

A brief history of the TOP program

The Dutch Transmural Developmental Support (TOP) program evolved directly from the evidence generated by the Stedelijk Transmuraal Interventie Programma voor Prematuur geboren kinderen en hun ouders (STIPP) study, a multicenter randomized controlled trial of the Infant Behavioral Assessment and Intervention Program (IBAIP) (66). This study demonstrated that a preventive home-based responsive parenting intervention for very preterm infants (< 32 weeks and/or < 1500 g) improved motor and cognitive development up to 5.5 years, enhanced maternal sensitivity, and reduced parenting stress, thereby providing a strong evidence base for further implementation (67-71). Building on these findings, the program was adapted and scaled up through a structured, stepwise implementation process. Bringing the TOP program to scale involved addressing both systemic and practical challenges (72). The nationwide implementation of the TOP program in the Netherlands serves as a model, highlighting strategies to transition from a research project into regular standard care covered by healthcare insurance.

Key components included: 1) Healthcare integration: Embedding interventions into routine healthcare services ensures sustainability and accessibility, as evidenced by the TOP program's reach to > 75% of the VP infants. In the Netherlands this translates to around 1400 children and families per year (73). 2) Standardized protocols: Developing a clear Theory of Change (TOC) and adaptable intervention protocols allow for consistent delivery while accommodating individual family needs. 3) Continuous Monitoring and Adaptation: Regular data collection on outcomes and evaluation through feedback from parents and TOP-interventionists, enables to improve the intervention and enhance impact.

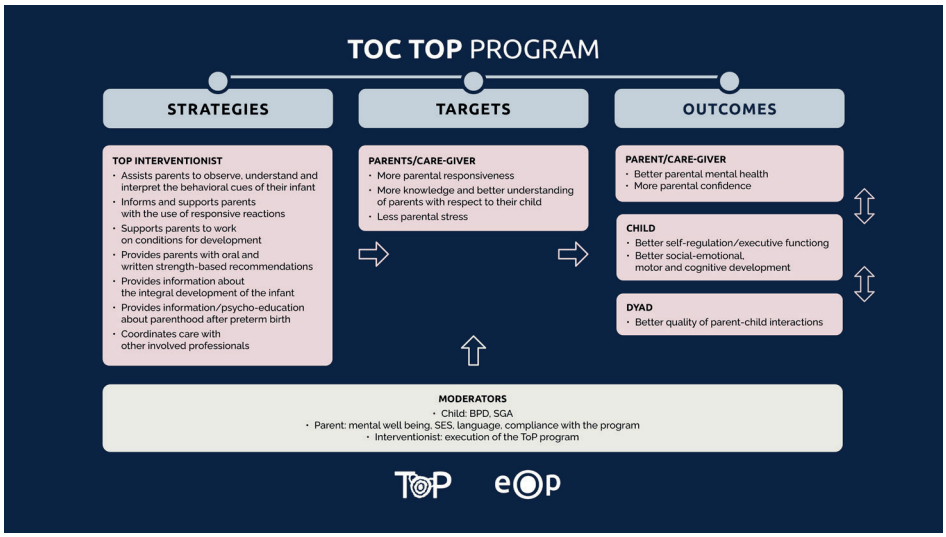

Figure 1.1. The Theory of change of the TOP program.
Abbreviations: BPD, Bronchopulmonary dysplasia; SGA, Small for gestational age; SES, Socioeconomic status.

Intervention protocol of the TOP program

The TOC underpinning the TOP program provides a structured dynamic framework that links its intervention strategies to targeted mechanisms of change and desired outcomes. The TOC is rooted in developmental science and aligned with the IDEAS Impact Framework™ of the Center on the Developing Child at Harvard University (74).

The TOC identifies how the TOP program's seven key strategies are expected to strengthen parental sensitivity, increase parental knowledge of infant behavior and development, and decrease parental stress (targets). Ultimately supporting the outcomes on three domains: 1) cognitive, motor, and socio-emotional development of PT infants; 2) improve parental confidence and mental health; and 3) improve the parent-infant interactions. Within this framework, moderators play a crucial role in shaping program impact. For the TOP program, important moderators include implementation fidelity, the degree to which interventionists adhere to and competently deliver the key strategies, as well as family factors such as socio-economic status (SES) and parental stress, and child characteristics such as being small for gestational age (SGA) and bronchopulmonary dyplasia (BPD).

The TOP program comprises twelve home visits, starting within the first weeks after discharge and continuing until the infant reaches one-year corrected

age (CA). Visits are scheduled every two to three weeks in the early months, with the frequency gradually decreasing as the child grows older. Each session lasts about 60–90 minutes and takes place in the family's home to provide support in the natural caregiving environment.

The TOP program is now delivered by 130 certified TOP interventionists. They are licensed pediatric physical therapists with the one-year additional specialized training in the theoretical background and practical strategies of the program. The intervention is guided by seven key strategies derived from the program's TOC. These strategies include: 1) observing and interpreting infant behavior and cues; 2) promoting sensitive and adequate parental responses; 3) creating conditions to foster optimal development; 4) informing parents about development and parenting; 5) collaboratively setting goals for the intervention; 6) actively intervening during parent–child interaction to model and coach responsive behavior; and 7) using a strength-based approach to empower parents.

During each visit, the interventionist observes both the infant and the parent-infant interaction, provides individualized feedback, and coaches parents in real time. Strength-based written recommendations are provided after each session, reinforcing positive behaviors and consolidating learning. Interventionists also coordinate care with other healthcare providers involved in the infant's follow-up, ensuring continuity and integration of services. Although the program does not follow a rigid protocol, it is built on a clear and consistent method. Flexibility is embedded to address the unique circumstances of each family. For example, the pace and emphasis of sessions may be adapted depending on infant characteristics, as well as family factors such as socioeconomic background or parental stress.

Adapt and innovate to improve the impact of the TOP program

The rapidly changing landscape of health care and the dynamic nature of scaling interventions requires evaluation, flexibility, adaptation, and innovation to remain responsive to the changing real-world health-care setting (75). A critical component in the successful implementation and scaling of early intervention programs, such as the Dutch post-discharge TOP program for VP infants, is the rigorous measurement of implementation fidelity. Fidelity ensures that interventions are delivered as intended, preserving the active ingredients responsible for positive outcomes (76, 77). Research has shown that programs implemented with

high fidelity, particularly in terms of adherence, dosage, and quality, can produce effect sizes two to three times greater than poorly implemented programs (78). These findings reinforce the importance of maintaining fidelity while allowing for strategic adaptations that align with the program's core principles and TOC.

Innovative adaptations to existing early parenting intervention programs have emerged as essential strategies to maximize the impact for long term neurodevelopmental outcomes of preterm born infants. Changes made by interventionists, to better fit the specific needs of the population, can lead to improved acceptability. However, these valuable unplanned modifications are often not registered or evaluated and therefore not implemented. To understand what, how, and when modifications are needed, and if they are fidelity-consistent, feasibility studies are recommended before efficacy testing or implementation and are therefore used for the research protocols in this thesis (79-81).

The process of adaptation and innovation can be guided by three key approaches: 1) improving execution quality through better training, coaching, and implementation monitoring; 2) expanding the target group to include infants with different risk profiles while ensuring the intervention remains developmentally appropriate; and 3) adding new features, such as accessible digital tools or tailored support for diverse cultural contexts.

Research-practice gap

Despite the maturity and nationwide implementation of the TOP program for VP infants, research-practice gaps remain. First, MLP infants (32–37 weeks gestation) represent most preterm births, however they are underrepresented in early intervention research (82). As a result, a gap has emerged in which parents but also clinicians, such as pediatricians and pediatric physical therapists, recognize the need for more structured follow-up and early intervention.

While the TOP program addresses the critical transition from hospital to home during the first year, evidence increasingly shows that developmental challenges do not resolve after infancy. VP children face ongoing risks in areas such as self-regulation, attention, motor coordination, and social-emotional development, which may only become apparent during the toddler and preschool years. At the same time, parents' needs also evolve, as they shift from navigating medical vulnerabilities to supporting autonomy, behavior, and learning. This creates research–practice gap: existing early interventions often concentrate on infancy, leaving a lack of structured, developmentally appropriate support for the later stages of early childhood. Staged, age-specific interventions are

therefore essential to provide continuity of care, respond flexibly to emerging challenges, and strengthen parental capacities across the child's developmental trajectory (83, 84).

Although parents of preterm infants often receive extensive medical and practical guidance in the neonatal period, their questions and concerns change as the infant grows and transitions into the home environment. They seek clear, timely, and personalized information on topics ranging from feeding and sleep regulation to long-term developmental outcomes yet often report fragmented communication and difficulties accessing trustworthy resources (85, 86). In this context, mobile health (mHealth) applications represent a promising development in healthcare, particularly for neonatal and preterm follow-up.

Research objectives

Post-discharge early intervention programs, delivered after hospital discharge, aim to improve the outcomes for preterm infants. When these programs scale up nationally, challenges arise in maintaining effectiveness, equity, and sustainability. This thesis builds on the existing body of knowledge from years of executing the TOP program and uses iterative design and pilot testing to investigate new approaches to improve the impact.

This thesis, comprising a series of interlinked studies, strategically reinforces three improvement areas: 1) improving execution quality of the intervention; 2) expanding the target group; and 3) adding new intervention strategies.

Specifically, it aims to:

1. Develop a fidelity tool for interventionists to ensure adherence and quality in intervention delivery.
2. Develop and evaluate a tailored TOP program for MP born infants.
3. Design and evaluate a booster intervention for VP born toddlers.
4. Examine the evolving information needs of VP parents during the first year after discharge.
5. Investigate the existence and quality of mobile apps providing post-discharge information for parents of PT infants.
6. Develop and evaluate the usability of an mHealth application to address parental needs for information in the first year after hospital discharge of their PT infant.

Contribution to practice and relevance for program evaluation

First, this research contributes to the improvement of the fully implemented TOP program. The aims of this thesis come from years of practice-based experience, specifically the input from parents, the Dutch parent association Care4Neo (87), TOP interventionists and other care professionals, and systematically collected care data by the Center of Expertise for Developmental Support of Premature Infants (EOP) (69). The purpose of the center is to develop and safeguard high-quality developmental support for prematurely born children by providing early intervention, training professionals, and advancing knowledge through research.

This thesis also contributes to the broader field of early childhood intervention and family-centered neonatal care by demonstrating how systematically integrating stakeholder perspectives and practice-based evidence can strengthen the impact, adaptation, and sustainability of intervention programs. By framing interventions not as static entities with strict protocols but as dynamic and adaptable systems, this work advances understanding of how theory-driven programs can evolve. In doing so, it underscores the relevance of continuous monitoring and innovation for ensuring that the benefits of early interventions reach diverse families and support the health and development of vulnerable infants.

Outline of this thesis

This thesis underscores the need for monitoring and innovation, ensuring that the benefits of early intervention reach all families equitably. This transformative approach positions the TOP program as a benchmark for future interventions, making a lasting contribution to the health and development of preterm infants globally.

Chapter 2 describes the development and evaluation of a Fidelity of Implementation Tool (FITT) to ensure consistent application of the TOP key strategies by objectively monitoring adherence and competence.

Chapter 3 describes the development and feasibility of the modified Dutch TOP program for moderate preterm born (MP) infants.

Chapter 4 describes the feasibility and potential effectiveness of an additional responsive parenting support for VP born infants at toddler age (18–24 months CA).

Chapter 5 aims to explore how parents of very preterm infants experience and articulate their need for information during the first year at home. A qualitative design was selected because it allowed for a deep understanding of lived experiences and Dutch context-specific needs.

Chapter 6 identifies and evaluates existing mobile information apps for parents of preterm infants targeting the post-discharge period. A systematic review design was chosen to provide a transparent and replicable overview of available apps across app stores.

Chapter 7 presents the user-centered design and usability evaluation of a digital information source (e-TOP) for PT parents during the first year at home after discharge.

Chapter 8 integrates in the general discussion and implications findings and highlights implications for practice, research, and policy.

This thesis not only enhances the theoretical understanding of how responsive parenting influences preterm infant development but also demonstrates how iterative innovation can transform into clinical care. By creating a feedback loop from scaled implementation back to innovation, the studies serve as a model for other interventions in neonatology and beyond. Its emphasis on usability, patient-centered innovation, scalability, and inclusivity redefines the scope of neonatal care, bridging gaps between clinical practice, digital health, and family-centered care in VP and MP infants and their parents.

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Chapter 2

Development and evaluation of a fidelity tool in a post-discharge responsive parenting intervention program for very preterm born children

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Abstract

Objective: The TOP program is a fully implemented responsive parenting intervention for very preterm born infants. Fidelity monitoring of interventions is important for preserving program adherence, impact outcomes and to make evidence-based adaptations. The aim of this study was to develop a fidelity tool for the TOP program following an iterative and co-creative process and subsequently evaluate the reliability of the tool.

Methods: Three consecutive phases were carried out. Phase I: Initial development and pilot testing two methods namely self-report and video-based observation. Phase II: Adaptations and refinements. Phase III: Evaluation of the psychometric properties of the tool based on 20 intervention videos rated by three experts.

Results: The interrater reliability of the adherence and competence subscales was good (ICC 0.81 to 0.84) and varied from moderate to excellent for specific items (ICC between 0.51 and 0.98). The FITT displayed a high correlation (Spearman's rho .79 to .82) between the subscales and total impression item.

Conclusion: The co-creative and iterative process resulted in a clinical useful and reliable tool for evaluating fidelity in the TOP program.

Innovation: This study offers insights in the practical steps in the development of a fidelity assessment tool which can be used by other intervention developers.

Introduction

Despite widespread agreement on the need for post discharge support for very preterm born (VPT) infants and their families, it is not often integrated into routine post-discharge clinical practice (1). In the Netherlands, a preventive home-based intervention program (the TOP program) for VPT children (< 32 weeks of gestational age and/or birth weight < 1500 g) and their parents is fully implemented, reimbursed by all Dutch Health insurance companies, and reaches yearly more than 75% of all Dutch VPT children (2). The TOP program is a process-oriented intervention with a theoretical framework that includes seven key strategies to target the desired outcomes (Figure 2.1; Theory of Change). The program is based on extensive research with sustained positive effects on infant cognitive, motor, and behavioral outcomes (3, 4) and has been gradually implemented.

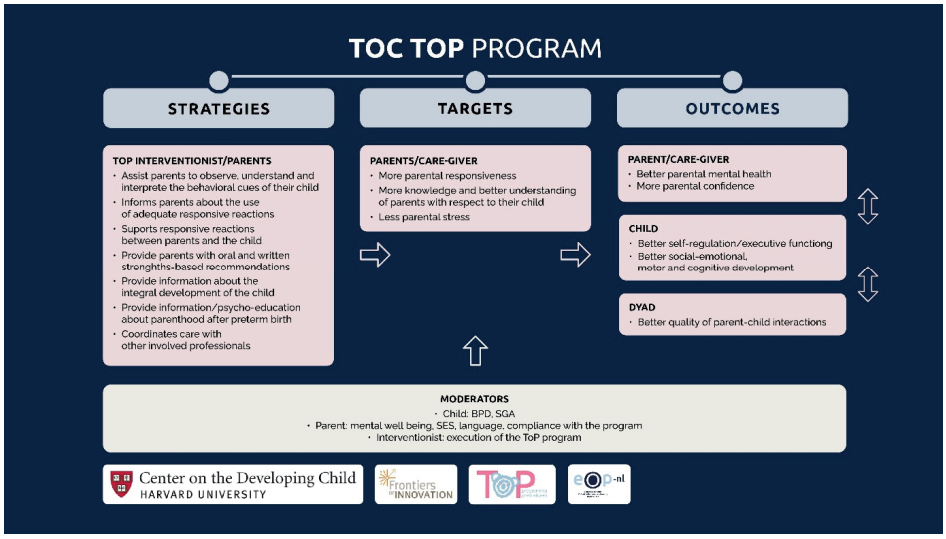

Figure 2.1. Theory of change of the TOP program.
Abbreviations: BPD, Bronchopulmonary dysplasia; SGA, Small for gestational age; SES, Socioeconomic status.

Scaling up an intervention from the controlled research conditions to routine care is long and complex process and requires selection of strategies relevant and feasible to that specific context (5). An important aspect during this journey is implementation fidelity; the degree to which the intervention is delivered as intended (6). Low fidelity could diminish the effect of interventions, but

adaptations that suit the needs and preferences of patients may also improve effectiveness. Although fidelity is associated with positive outcomes, it may be difficult to achieve in routine care and sustaining program fidelity of large-scale disseminated interventions has rarely been studied systematically (7-9).

Fidelity assessments are especially important to use for interventions where multiple factors can influence the fidelity of the intervention delivery, such as heterogeneity of interventionists, participant characteristics, and family circumstances (10). In order to evaluate the fidelity of the TOP program we first needed to develop an intervention-specific fidelity assessment tool. Despite the presence of validated fidelity tools for several parent-child interventions, these can only serve as examples as the target groups and the intervention strategies differ from the TOP program (11-17). There is a great diversity in concepts and constructs for establishing fidelity and fidelity measures used for parenting programs differ from observational methods such as live observations, video-recorded sessions and non-observational measures such as self-report (18, 19). An (2020) described a useful multidimensional construct for intervention fidelity, including adherence to key components, quality of intervention delivery, amount of intervention delivered, participant responsiveness, and program differentiation. However, guidance on the processes involved in developing a feasible and valid fidelity tool within complex interventions at scale is limited (20, 21).

The TOP program is part of an interdisciplinary learning community created by the Center on the Developing Child at Harvard University. Their IDEAS Impact Framework™ provides an innovative approach for program development and evaluation to improve outcomes (22). During the implementation of the TOP program, the four guiding principles: precision, fast-cycle iteration, co-creation, and shared learning, have been adopted (23). Starting with the precision, a Theory of Change of the TOP program was developed, a framework that links interventions strategies, via targets, to the outcomes.

The next step is to include a fidelity measure that is closely tied to our intervention strategies and training materials. Following an iterative process enables rapid learning and making refinements while developing the tool. A partnership between researchers, developers, educators and interventionists contributes to the development of a realistic and practicable tool. This increases the likelihood that the tool has both content validity and is feasible for use in practice. This study aimed to develop an intervention-specific fidelity tool, in co-creation with stakeholders, and study the psychometric properties to be able to guarantee the quality of the scaled-up TOP program.

Methods

This study consisted of three distinct phases. In phase I, a draft of a fidelity tool was developed and tested in a co-creation group. The co-creation group consisted of five certified TOP interventionists with more than two years of experience in the execution of the TOP program and three members of the research team. The results of the pilot testing in phase I gave rise to an extensive evaluation and necessary adaptations. In phase II, the co-creation group evaluated in two work sessions the results of the pilot testing and user experiences. Recommendations for improvements were formulated and concerned the rating method, the item content, and training of expert raters. The adaptations led to the Fidelity of Implementation Tool TOP program (FITT). In phase III, the psychometric properties of the FITT were studied.

The Medical Ethical Review Committee (METC) of the Academic Medical Center Amsterdam provided a waiver for ethical review of this research. Parents signed informed consent for collecting personal data, video recording, and self-report evaluations following the Privacy Act. In addition, the TOP interventionists signed informed consent for being recorded.

Participants

Families who participated in the TOP program with a VPT child (< 32 weeks of gestation) or with very low birth weight (< 1500 g) were eligible for both the pilot- and psychometric evaluation study. Sociodemographic and perinatal factors of included participants are presented in Table 2.1. We classified the children in an even distribution of child's corrected age (CA) since the deployment of intervention strategies could differ per developmental stage and the course of the 1-year TOP program.

Tool development

Phase I: Development and pilot testing

The first author (MF) developed a preliminary tool based of two scales: 1) adherence to program content and 2) competence in program delivery. Key intervention strategies from the Theory of Change (TOC) (Figure 2.1) were operationalized into observable items for the Adherence scale. The techniques to enhance the execution of the key TOP strategies were operationalized for the Competence scale. Two key strategies of the TOP program ('written strength-

based recommendations' and 'coordinates care with other involved professionals') took place outside the home visit and were therefore not included in the tool. Ratings were based on the number of opportunities the interventionist utilized to apply the described key strategy or used a competence technique. At the same time, missed opportunities led to a lower score on that item. Two methods were explored for measuring the fidelity, namely a self-monitoring- and a video-based observation tool. The preliminary draft of the measuring tools was discussed,

Table 2.1: Participant characteristics

	Total group (n = 19 families)
Perinatal factors	
Gestational age, n	
< 28 weeks	2
28–32 weeks	15
≥ 32 weeks	2
Birth weight (gram), M (SD)	1419 (344)
Days in hospital, M (SD)	53 (23)
Gender child boy, n	11
Singleton, n	19
Age during the home visit (CA), n	
0–3 months	3
3–6 months	9
> 6 months	8
Social factors	
Age mother, years, M (SD)	34 (5.8)
Age father, years, M (SD)	35 (7.5)
Mother born in the Netherlands, n	15
Father born in the Netherlands, n	16
Dutch speaking family, n	16
Marital state married/together, n	17
Level of education[a] - Mother, n	
Low	1
Middle	9
High	9
Level of education[a] - Father, n	
Low	1
Middle	8
High	9
Missing	1
No. home visits (median, range)	5.5 (3–11)
Number of visits with both parents, n	4

There were 20 videos available from 19 families.
[a] Low = primary school, pre-vocational secondary education; middle = senior general secondary, education, pre-university, or secondary vocational education; high = higher professional education or university education.

clarified, and refined in the first co-creation session. Then, video-recorded intervention sessions were used to discuss the items and response options critically. In between sessions, the TOP interventionists reflected on how well the FITT draft matched their actual intervention execution after their regular TOP home visits. In the following co-creation sessions, all items were further discussed to determine if they should be retained, modified, or missed core components.

The adherence scale was operationalized in eleven items that measured the frequency of applying the described key strategies, on a 3-point rating scale. For the competence scale, four items that represented the strength-based approach and skills to transfer information to the parents were identified, rated on a 3-point scale. A total impression item was added to assess the overall quality of the intervention and was scored on a 10-point Likert scale with a score ranging from 1 (very low) to 10 (excellent). In addition, the therapists indicated the need to outline family characteristics that may influence parental engagement. Therefore, the item "Parental availability" was added, measuring how receptive or engaged the parent was during the intervention session, also rated on a 3-point scale. The manual, including detailed descriptions and illustrative examples, guidelines for scoring, and decision rules, was created simultaneously.

Procedures pilot testing

Phase I: Pilot testing

TOP interventionists (n = 5) were video recorded during two of their routine TOP home visits. In addition, they filled in the self-report after ten of their TOP home visits, including the two video-recorded home visits. An independent researcher (EV) compressed the recorded home visit (1 h) to capture the intervention's core in about 25 min. The five TOP interventionists rated the video-recorded home visits (n = 8), except their own, to assess the inter-rater reliability. The first author (MF) rated the complete video-recorded home visits. The self-reported scoring of the raters was compared with the rating of the first author.

Phase II: Adaptations and refinement

In two work sessions, the co-creation group evaluated the results of the pilot testing and user experiences. Regarding the video-based rating, all individual items were discussed using the comments and reflection notes from the participating TOP interventionists. In addition, the rating system was discussed, and the reliability of the video rating by TOP interventionists as raters was evaluated.

The research team changed and clarified definitions per item and tested the adaptations with training videos.

Phase III: Psychometric evaluation of the FITT

The FITT contains four items for the adherence scale and five items for the competence scale, a global impression score (item 10), and an item on parental availability (item 11.1 and 11.2). Items 1–9 and 11 (Parental involvement during the home visit) are rated on a 3-point Likert scale. Each score per item is described comprehensively and contains examples (see Appendix 2.2), and scores 1–3 indicate ascending quality of the execution. Item 10 is the global impression score on a scale of 1 (very bad impression) to 10 (excellent impression).

Three TOP lecturers, assigned as expert raters, evaluated twenty regular TOP home visits videos. The collected videos were reviewed for sufficient quality and diversity by one researcher (EV). Just as in phase I, complete video recorded home sessions were compressed into 20–30 min videos to capture the intervention's core. The manual with guidelines for the use of the FITT, description of the FITT items with examples and scoring criteria accompanied the video vignettes. Rating of the items was performed after the first observation of the video vignette, and the final scoring was given after the second viewing.

Statistical analyses

A dataset was created in Castor, a web-based system to build electronic Case Report Forms (CRF). Subsequently, the data was exported to Statistical Package for the Social Sciences (SPSS, version 26.0).

In phase I, the agreement between self-report and expert video rating for the composite scores of the subscales were measured with a two-way random effects model, single measures (24). Cohen's Kappa with quadratic weighting (25) was calculated for individual items of the FITT. Interrater reliability of the video ratings was measured with Krippendorf's alpha (Kalpha). The Kalpha was chosen because each video had missing values, as the TOP interventionist did not rate their intervention sessions. Furthermore, the Kalpha does not require minimal sample sizes (26). In phase III, the inter-rater reliability of the subscales and the individual items was measured with the ICC (two-way mixed model, mean-rating, absolute agreement) with a 95% confidence interval. Values of the ICC were described as poor (< 0.50), moderate (0.50–0.75), good (0.75–0.90) or excellent (> 0.90) (27). Chronbach's alpha (α) was used to measure the internal consistency of the subscale's adherence and competence. The Spearman's Rho

(r_s) was calculated to examine correlations between the adherence and competence scales and both to the total impression score.

Results

Phase I

The agreement between the self-report (n = 10) from the five interventionists and expert video rating was below the acceptable level of agreement (ICC < 0.5) for individual items and the composite scales. The inter-rater reliability for individual items measured with the Kalpha was poor for the individual items and the two subscales.

Phase II

The evaluation led to necessary adaptations (see Appendix 2.1). Recommendations for improvements were formulated and concerned the 1) method of rating; 2) item content; and 3) raters.

All five TOP interventionists agreed that recalling the details of the home visit was too difficult. Since the rating scale was based on the number of used opportunities, the lack of awareness or recalling missed opportunities led to possible over-estimation of the used strategies. The context of the session, family circumstance, and prior interventions sessions played a role when filling out the self-report, also causing subjective and more positive assessments by the interventionist. Therefore, the development of the self-report fidelity tool was stopped. Furthermore, instead of rating the number of used opportunities, it was decided to provide all items with specific objective criteria to represent the particular level of quality Items were scored on a Likert scale (0 = No or insufficient execution of the strategy/competence, 1 = sufficient execution of the strategy/competence, 2 = good execution of the strategy/competence. Additional guidelines, explanations, and examples for scoring per item were added to the manual.

Regarding item content, changes and clarifying definitions per item were made, and some competence items were added and adapted to improve the competence scale of the tool further (see Appendix 2.1). Finally, the video rating by interventionists as raters was evaluated. Although the raters worked closely on developing the scale, deviant interpretation of the items and no additional training in the use of the tool may have caused the poor inter-rater reliability. Another aspect, rating fellow co-workers could have counted for higher ratings.

Extensive training of fidelity raters in this phase of the iterative and flexible development process was not considered efficient. Therefore, the three experienced lecturers with additional training in rating videos were assigned as expert-raters for phase III.

Phase III

Interrater reliability from the three expert raters across the 20 videos for both subscales and total impression score were excellent, with average measures ICC > 0.80 [95% CI 0.60 to 0.95, p < .001] (Table 2.2).

For the individual items, the inter-rater reliability was substantial; nine items scored ICC values > 0.60 [95% CI 0.62 to 0.99]. Only item 4 (Informing about development and parenting) of the adherence scale and item 9 (Use of didactic skills) of the competence scale had ICC scores < 0.60 [range 0.51 to 0.56] (Table 2.2). The internal consistency as measured by Chronbach's alpha (α) was .57 for the adherence scale and .77 for the competence scale. The associations between the subscales and each subscale to the total impression scale were (very) strong with Spearman's rho ranging between r_s 0.70 and 0.82.

Table 2.2. Results interrater reliability of the FITT (item and composite scores)

	ICC	95% CI
Item 1 – Reading behavioral cues	0.645	0.271–0.846
Item 2 – Promoting adequate responsive reactions	0.852	0.690–0.937
Item 3 – Creating conditions for enhancing development	0.617	0.062–0.779
Item 4 – Informing about development and parenting	0.510	0.122–0.822
Item 5 – Discussing intervention goals	0.804	0.588–0.916
Item 6 – Intervening in parent-child interaction	0.720	0.422–0.885
Item 7 – Using strength-based approach	0.635	0.258–0.841
Item 8 – Timing and dosing in information transfer	0.811	0.607–0.919
Item 9 – Use of didactic skills	0.562	0.127–0.808
Item 11.1 – Availability of the mother/parent 1	0.616	0.213–0.834
Item 11.2 – Availability of the father/parent 2 Composite scores and global impression	0.986	0.971–0.994
Adherence scale	0.808	0.601–0.917
Competence scale	0.842	0.666–0.933
Total impression score	0.877	0.764–0.952

ICC estimates were based on a mean-rating (k = 3), absolute-agreement, two-way mixed-effects model; Values of the ICC were described as poor (< 0.50), moderate (0.50–0.75), good (0.75–0.90), or excellent (> 0.90).

Discussion

This study describes the process of developing and evaluating an intervention-specific fidelity measure for the implemented TOP program for VPT and their parents. The co-creative and iterative process led to a reliable tool that objectifies intervention fidelity and is relevant for adding transparency and transferability to the TOP program.

Self-report vs. video-based observation

Initial evaluation of the reliability in phase I was not very promising. The results showed subjective bias in the self-report: higher fidelity ratings compared to the observational measure. The interventionists agreed that scoring retrospectively and objectively recalling details with the fidelity checklist was too difficult. Although this systematic bias in fidelity self-reports is confirmed by other studies, the self-report method would have been less complicated and costly in an integrated routine post-discharge intervention and was therefore explored (10, 12, 28).

The drawback of the video observations with high labor costs, time involved to train raters and the challenges to recruit families and interventionists outweighed the rich and unique source of information coming from video observations of regular TOP home visits. The results of this study support the use of videotaped interventions as a meaningful method for fidelity assessment (29).

Translating the key strategies and competent execution into observable items

The TOP program is a complex intervention since it contains several interacting components and interventionist have to tailor the intervention to the family context to carry out the key strategies of the program. We wanted to assure that the fidelity tool captured the adherence to the key strategies as described in the Theory of Change (Figure 2.1) and the competence in program delivery following the described pedagogical approach and vision of the program. However, the more detailed and subdivided elaboration of the key strategies in phase I led to substantive interpretation issues. The level of detail was reduced and described in more observable behavior to leave less room for ambiguity in interpretation and scoring. The number of items representing the key strategies was reduced to make the tool applicable for future purposes.

Since each item in the FITT represents a different key strategy or meaningful competence, we aimed for ICC values > 0.50 at the item level. Two items (4, 9) only just complied with the ICC criteria and might need better descriptions and examples. The interrater reliability of the Adherence and Competence scales and the total impression scale scored in the excellent range. Composed scores usually demonstrate stronger reliability than individual items. (16) The good agreement between the subscales and each subscale to the total impression scale indicates that these FITT scores can be used to establish insight in the intervention fidelity. By relying on a single video observation to rate the fidelity of a 1-year process-oriented intervention we could risk making erroneous conclusions on the fidelity, as also described by Cross et al. (30). Which number of home visits should be rated with the FITT to obtain insight into the overall quality of the execution of the intervention needs to be determined in future research.

Process of development

We partnered with TOP interventionists, researchers and program developers to develop a fidelity monitoring tool that would capture the essential elements of the TOP program as described in the TOC of the program and also matched the execution of regular TOP home visits. Using an iterative approach, we employed concepts and made several adaptations and tested several options using video vignettes from home visits. To distill a complex intervention into a practical number of measurable items and for the tool be readily available for later use we recommend this partnership between researchers, educators and practitioners. Although we used an iterative process and did not determine the needed phases in advance, creating the fidelity tool ended in a 3-step process. Practitioners are very sensitive to the challenges of fellow interventionists, and we would therefore recommend independent raters and extensive training for practitioners. Forgatch also described this bias and suggested raters that are unfamiliar with the trainee (31).

Limitations and future research directions

In this study, we addressed and contributed to the gap in fidelity measurement tools for interventions at scale. We developed the fidelity tool to measure two constructs; adherence with the delivery of the key strategies and competence in intervention delivery and added scores for parent availability and total impression. However, to measure the full construct of intervention fidelity and relate this to intervention outcomes, the other constructs such as dose (amount

of intervention delivered) need to be included as well (32). Since the actual delivery of the home visits is registered, this can easily be added. For measuring parental responsiveness, we used one overall score (item 11) to describe the parental engagement with the interventionist and active participation in the interaction with their child. Since parental responsiveness is a target in our program and all of the key strategies and intervention specific competences are designed to let the parents be in the lead and strengthen parent-child interactions parental responsiveness can vary and improve during the trajectory and should be measured more than once.

A limitation that may have affected our reliability results is the restrictive 3-point Likert scale. Although this seemed to be the optimum number of response categories in the development of the tool, it caused lower variance in certain items while coding the videos. ICC values are likely to have been reduced due to the restriction of range in scores. Availability of more diverse videos with various distribution in scores could have resolved this issue.

Future research directions

The criteria to determine what an acceptable level of fidelity is, still needs to be determined. This is an important next step when systematically examining the associations between program fidelity and outcomes. The TOP program has been carefully implemented but may need adaptations in response to changes in participant needs, new insights, and other resources available. The FITT has great value to support further development of the intervention and enlarge its impact since it allows to monitor the pre-defined changes.

Conclusion

This study describes the iterative development of the FITT, an intervention-specific fidelity measure for the TOP program. The co-creative and iterative process led to a tool that objectifies what is done in the home visits, thereby adding transparency and transferability to the TOP program. The high association between the total impression item and subscales shows that the FITT identifies and captures the unique execution of the TOP intervention. By working together with practitioners, researchers, and lecturers, the FITT became a relevant and reliable fidelity tool and will serve for future evaluation of the TOP program and contribute to measuring the professional development of the TOP interventionist and the continuous quality improvement efforts. We would strongly

recommend that when a new intervention is developed, a fidelity measure should be developed simultaneously to improve the study's internal validity and safeguard the quality of the intervention during the implementation process.

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Appendix 2.1. Revisions after pilot testing and description FITT items phase III

Pilot FITT item	Problems with the pilot item	Adaptations to current item	Current item	Description current FITT item
1. TOP therapist helps the parent to observe and interpret child's behavioral cues **2. TOP therapist provides the parent with information about behavioral cues that indicate disbalance and/or stress** **4. TOP therapist provides parents with information about self-regulatory behavioral cues** **6. TOP therapist gives information about behavioral cues indicating approach and exploration**	- Item 1, 2, 4 and 6 were separate items of the same key strategy and were too detailed for scoring - Scoring was unclear regarding the frequency and completeness of the execution of the strategy and when parents already reacted sensitive and only confirmation of parental behavior is appropriate - Raters had different views on description "providing information"	- These items were merged to current item 1, reflecting key strategy 1 (TOC) - Scoring guidelines were re-defined - More detailed description and diverse examples of item 1 were given in the instruction manual - Clarification of "providing information" was given for this item and distinguished with providing information included in other items	**1**	The TOP therapist names child's behavioral cues and gives information about the interpretation of the behavioral cues
3. TOP therapist facilitates parental responsive reaction: support comfort and safety **5. TOP therapist facilitates responsive reaction: co-regulation** **7. TOP therapist facilitates responsive reactions: adding information/scaffolding**	- Item 3, 5 and 7 were separate items of the same TOC key strategy and too detailed for scoring - Scoring was unclear, since "facilitating" can be done in different ways (verbal cues, practical guidance)	- These items were merged to current item 2, reflecting key strategy 2 and 3 (TOC) - The term "facilitate" was replaced with the term "support" and a more detailed description of the interpretation was added - Rating of the use of various facilitating techniques by the observed therapist was transferred to a new competence item 9	**2**	The TOP therapist supports the parent to apply the adequate responsive reaction

Appendix 2.1 continues on next page.

Appendix 2.1. *Continued*

Pilot FITT item	Problems with the pilot item	Adaptations to current item	Current item	Description current FITT item
		- This item was added because in the TOP program there is attention for delays in motor development which can cause stagnation in other developmental areas. Additional exercises can be indicated and carried out by the TOP therapist.	**3**	TOP therapist supports parent in creating conditions to enhance development
9. TOP interventionist provides information about current developmental stage and gives relevant information about next stage **10. TOP therapist gives information about impact preterm birth on parenthood**	- These items represent the key strategies 5 and 6 from the TOC and needed more detailed scoring guidelines - Raters had different views on description "providing information"	- Item 9 and 10 were combined - Clarification of "providing information" was given for this item and distinguished with providing information included in other items - The possibility to check off the addressed developmental domains was added	**4**	The TOP therapist informs the parent about developmental domains and parental well-being after preterm birth
11. TOP therapist aligns care plan with parents and other caregivers	- This item was related to an important TOC strategy 7 but not applicable within every home visits	- Item was moved to the additional checklist in order to keep the FITT suitable for every session		
12. TOP therapist reacts responsively to parents and their circumstance	- Description and scoring was unclear - To be effective and structure the sessions, intervention goals for the home visit needed to be set explicitly and TOP therapist must use and create situations to work in the actual parent-child interaction	- This item was translated to new item 5 and 6 with detailed descriptions	**5** **6**	The TOP therapist formulates intervention goals for each home visit The TOP therapist uses or creates situations to work in the actual parent-child interaction

Pilot FITT item	Problems with the pilot item	Adaptations to current item	Current item	Description current FITT item
8. TOP therapist uses specific and strength-based recommendations	- Item needed clarification - Not a strategy, but should be scored as a competence item	- The strength based approach was clarified and seven observable aspects were defined - This item was transferred to the competence scale	**7**	The TOP therapist works according the strength-based approach
13. TOP therapist transfers information and knowledge correctly and adequate	- It was unclear how to deal with fragments where the therapists uses jargon or clearly explains too much and interferes with parent-child interaction	- This item was translated to the new competence item 8 which reflects the timing and dosing of information during the intervention session - Aspects of this item 13 were incorporated in the scoring guidelines of the new item 4 (e.g. adequacy of information)	**8**	The TOP therapist uses good timing and dosage in the information transfer to parents
14. TOP therapist uses verbal skills to improve responsive parenting **15. TOP therapist uses active learning skills to improve responsive parenting**	- Item description of item 14 and 15 and rating was not clear - Item 14 en 15 were related	- Didactic skills were better defined, combined and examples were added in the manual - Also for a correct execution of this competence, parent's response was included in the scoring guidelines	**9**	The TOP therapist uses didactic skills for the transfer of information
Total impression	- Subjective item, no problems	- Description clarified in: Total impression of the entire intervention session	**10**	Total impression of the entire intervention session
Availability parent (s)	- The manual was unclear how to deal with fragments in which two parents are present but not both involved	- Description was clarified - Not applicable was added if other parent was not present	**11.1** **11.2**	The availability of the parent(s) involved in the intervention

Appendix 2.2: Examples of operationalization adherence item 1 and competence item 7

Item 1: The TOP interventionist names the child's behavioral cues and gives information about the interpretation of the behavior

This item represents key strategy 1 of the TOC; Assist parents in observing, understanding, and interpreting their child's behavioral cues. Behavioral cues are classified in different behavioral systems; autonomic, motoric, state, and attention/interaction.

The interpretation of the behavioral cues gives meaning to the child's goals. We interpret the behavior according to the categories used in the TOP program, approaching behavior, regulation behavior, or dysbalance/stress behavior. For this item, the provision of adequate information of the TOP therapist counts. If the interventionist occasionally uses a misinterpretation and adapts or corrects, this does not alter the rating since this can be informative for parents.

ITEM 1: Scoring guidelines

Score 0: No or insufficient use of the strategy (item 1)

Criteria:

- The interventionist does not use the key strategy or does not give accurate information, or
- The interventionist only mentions single behavioral cues, or
- The interventionist only gives interpretation separate from behavioral cues.

Score 1: Sufficient use of the strategy (item 1)

Criteria:

- The interventionist names only single behavioral cues linked to the interpretation.

Score 2: Good use of the strategy (item 1)

Criteria:

- The interventionist names behavioral cues linked to the interpretation in more than one behavioral system or more behavioral cues in the same behavioral system.

Examples scoring item 1

Score 0
"Did you see that he places his feet together."
"He seeks support with his feet against the edge of his crib."

Or confirming parents:

The parent mentioned, "*I see in his eyes that he is tired*". The therapist's reaction: "*you can bring him to bed if you think he is tired*". This is scored as insufficient use of the strategy because the interventionist does not mention concrete behavioral signs and only confirms the interpretation.

Score 1
"He is attentive to his toys even when they roll out of sight. It looks like he would like to try to get them again."

Or confirming:

"What do you know his behavior well? You describe that he is rubbing his eyes, and this is how you see he is getting tired."

Score 2
"Now he is on his tummy; you can see he is turning red; he groans and grumbles. It looks a bit too strenuous at this moment, do you think that also?"

Or confirming:

"He takes your hand with his finger and focuses on your face while he is drinking his bottle. Because you give him so much support and he can hold on to your finger, he is successful in coordinating his sucking, breathing, and swallowing."

Item 7: The interventionist works according to the strength-based approach

This item is about the extent to which the interventionist uses the strength-based approach in the TOP intervention. Besides using the approach in the actual intervention, this also has a concrete application in the parent report given after each intervention session. Elements required for the parent report are Positive, Rationale, Functional, Open, and Individual.

The child and the parent's protective factors, possibilities, and developmental chances are the core of this approach and form the port of entry in the TOP intervention. The strength-based approach adds to parents' feelings of competence and regards the parents as experts. Enhancing parental feelings of confidence and joy in the interaction ensures parental responsiveness that is more consistent.

The strength-based approach is audible in the information transfer during the home visit and scored with the following aspects:

a. The interventionist formulates positive and non-judgmental.
b. The interventionist gives open suggestions and recommendations that support the responsive relationship or development.
c. The interventionist connects to the child's possibilities and works from the strengths of the child.
d. The interventionist connects to the support the parent can provide at that moment.
e. The interventionist is confirming in the communication about the parental efforts.
f. The interventionist has an open attitude towards parental upbringing.
g. The interventionist uses the ideas and suggestions from the parent during the intervention.

The strength-based approach is different from the more traditional problem-based approach and norm-based testing for delays. The TOP program is a preventive program, and early detection of problems or delays is essential and additional support (key strategy 7) or treatment (item 3) will be applied if indicated.

ITEM 7: Scoring guidelines

Score 0: No or insufficient execution of the TOP competence

Criteria:

- The interventionist does not work according to the strength-based approach or
- The interventionist uses only 1 or 2 aspects (a-e).

Score 1: Sufficient execution of the TOP competence

Criteria:

- The interventionist uses at least three aspects (a-e).

Score 2: Good execution of the TOP competence

Criteria:

- The interventionist uses all described aspects (a-e), and
- The interventionist has an open attitude towards the parental upbringing, and
- The interventionist uses the ideas and suggestions from the parent during the intervention.

Chapter 3

Feasibility of a Dutch post-discharge parenting intervention (TOP program) for moderate preterm born infants

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Abstract

Background and aim: Moderate preterm (MP) birth is associated with an increased risk of developmental problems. However, post-discharge support for this group is scarce. The aim of this study was to evaluate the feasibility of a post-discharge parenting program (TOP program) for MP infants. Three feasibility dimensions were evaluated (1) recruitment capability and compliance, (2) intervention acceptability, and (3) limited efficacy testing.

Methods: A group of MP infants with a gestational age (GA) between $32^{0/7}$–$34^{6/7}$ weeks and their parents received six home visits by a TOP interventionist until 6 months corrected age (CA). A pre-posttest intervention design with quantitative and qualitative measures was used. Recruitment capability and compliance, acceptability, and satisfaction with the intervention were evaluated using a questionnaire, checklists, interviews, and a focus group. Infant socio-emotional development, parental distress, self-efficacy, and reflective functioning were measured with questionnaires. Observation measurements were used for infant motor development and parental sensitivity.

Results: Thirty-two families completed the six home visits. The satisfaction rate (scale 0–10) was remarkably high (Mean 9.4, range: 8–10). Parents reported that the program was suitable, enhanced their understanding of their infants' developmental needs, and increased their self-efficacy. The infants showed age-appropriate motor and socio-emotional development post-intervention. Parental self-efficacy, reflective functioning, and sensitivity improved from pre to post intervention, with small to large effect sizes.

Conclusion: The study demonstrated high compliance, acceptability, and satisfaction with the TOP program for MP infants with promising infant and parent outcomes. This study contributes to the preparatory work prior to a larger scale evaluation and dissemination.

Introduction

Preterm birth can adversely affect the developmental outcomes and quality of life of the child. Although the risk of severe infant morbidity, such as developmental disabilities, declines with increasing gestational age (GA) (1, 2), children born with a GA between 32 and 37 weeks, defined as moderate and late preterm (MLPT), have more hospital re-admissions than full term (FT) infants in the first thirty days after discharge for e.g. jaundice, respiratory distress, crying, and vomiting (3). They also have a higher risk of feeding problems (4), language problems (5), impaired neuro-psychological functioning, emotional and behavioral problems (6, 7) and poorer academic performances than FT infants (8, 9). In later life, MLPT birth even increases the risk for mental health disorders, obesity, and coronary artery disease (10, 11). MLPT birth can also negatively impact parents, as demonstrated by increased parental stress (12, 13), lower confidence (14), and increased risk for post-partum depression (15). This can in turn negatively affect parent-infant co-regulation and parenting behavior (16). The quality of caregiving is central to the child's developing resilience in the face of adversity, however, parenting a preterm infant can be challenging as preterm born infants show more diffuse behavioral signs, less approach behavior and more regulation difficulties (17). Being able to understand the behavioral signs, interpretation of the infant's needs and being able to co-regulate are connected to parental self-efficacy and reflective functioning and have emerged as an important clinical target for interventions. Parental self-efficacy, first described in 1977 as parents' belief in their ability to successfully parent their child (18), is considered to be important for the health and well-being of parents and influences infant regulation and parents' perception of their infant's temperament (19, 20). Parental reflective functioning (PRF), described as the ability of parents to mentalize and adjust to the mental needs of their infant, is also considered to affect infant development, and is especially important for preterm infants given the challenges they face (12).

Early intervention incorporating multiple strategies, active parental participation, and focus on the infant, parents or parent-child interaction could reduce the risk of adverse outcomes and enhance infants' development (21, 22). Despite increased risks of adverse outcomes for MLPT infants, they do not routinely enroll in specialized medical follow-up or early intervention programs (23).

Low health literacy (LHL) is associated with poorer health outcomes, including higher rates of hospitalization and suboptimal use of preventive services.

For parents of preterm infants, this can translate to difficulties in managing the infant's health needs, potentially leading to worse developmental outcomes (24). We aimed to include a relatively high number of parents with a low educational level in this study to learn whether the transfer of information during the home-visits or the additional e-TOP app was suitable form them. In the Netherlands, the evidence-based TOP program for very preterm (VP) children (GA < 32 weeks and/or birth weight < 1500 g), carried out by pediatric physical therapists (TOP interventionists), is implemented as usual care (25). The TOP program contains seven strategies to strengthen parental responsiveness, improve parents' knowledge and understanding of their child's developmental needs, and decrease parental stress (Figure 3.1). We hypothesized that the strategies of the TOP program are also suitable to address the challenges MP infants, and their parents face after being discharged home and can improve developmental outcomes.

To determine whether an intervention can be expanded to a different population, feasibility studies are recommended before efficacy testing or implementation (26, 27). We were aiming to answer the research questions: 1) recruitment capability and compliance. Will MP parents be identified by hospital staff. Will MP parents participate and complete the intervention? 2) intervention acceptability. Do parents perceive the adapted TOP program as appropriate, and does it accommodate their needs? 3) limited-efficacy testing. Are the measures appropriate and sensitive to evaluate change, is there preliminary evidence for positive changes. The aim of this study is to evaluate if the modified TOP program is feasible for MP infants and their parents.

Methods

Setting and procedure

The study was conducted by the research team, consisting of researchers and educators from the Dutch Expertise Centrum Ontwikkelings ondersteuning Prematuren (EOP), affiliated with the Amsterdam Medical Center, and the Centre of Expertise, Faculty of Health, Amsterdam University of Applied Sciences The EOP is responsible for coordination and implementation of the existing TOP program for very preterm born infants. Prior to the enrollment of participants, the research team recruited fifteen affiliated and experienced TOP-interventionists, from different geographic and socio-economic regions. The TOP-interventionists all had a working relationship with the hospital staff and provided the hospitals with information about this feasibility study.

The feasibility of the adapted TOP program was assessed in a group of MP infants and their parents, using pre and post intervention measures. Enrollment for the study was from November 2022 till March 2023. Eligible participants were approached by the hospital staff (pediatrician or pediatric physical therapists) of the participating secondary-care level hospitals. When interested, participants received an information letter including a QR code referring to a short video-clip about the study The TOP-interventionist in that region was informed by the hospital staff. The TOP-interventionist was the primary contact for the parents and scheduled the pre-intervention home visit and also informed the study center. During the pre-intervention home visit, the TOP-interventionist obtained written informed consent (IC). Additional written consent was asked for filming of the parent-child interaction. When provided, this interaction was filmed during the same home visit. All participants that provided consent received the adapted TOP-program in addition to care as usual. Non-participants received care as usual, which was not provided by TOP-interventionists. In case of non-participation after the pre-home visit, the first author collected additional information by making a phone call to TOP interventionists to elucidate the parents' concerns.

After the study center received the signed IC, both parents received an e-mail with the invitation to complete online questionnaires at baseline (T0). One reminder was sent to the families via e-mail or WhatsApp. Perinatal variables (GA, sex, multiple birth, birth weight, APGAR score, length of hospital stay, and morbidities) were extracted from the medical record by the referring hospital staff and send to the TOP-interventionist. The TOP interventionist transferred this data to the study center using a system for securely sending data.

During the last home visit, parent-child interaction was videotaped again. After completion of the intervention, parents again received an intervention to complete a set of online questionnaires. The first author also interviewed several parents about their experiences with the intervention. After completion of all home visits, TOP interventionists participated in a focus group on their experiences with the execution of the intervention and participating in the study.

The study protocol was approved by the medical Ethical Committee of the Amsterdam UMC (NL78996.018.21).

Intervention development and training

The intervention protocol for MP infants was based on the existing evidence-based TOP program. This post-discharge intervention for VP infants consists of 12 home visits by a TOP interventionist in the first year. In co-creation with

parents, TOP interventionists, pediatric physical therapists (PPT), and pediatricians, adaptations to the TOP protocol were established. An important adaptation was to shorten the TOP program for MP infants to six home intervention sessions until the corrected age (CA) of 6 months. Each home visit had a duration between 45 and 60 min.

The use of the seven key strategies including the parent-report with individualized strength-based recommendations were indicated as essential and remained unchanged (Figure 3.1) (30). To strengthen two key strategies (providing information about the integral development of the child, parenthood after preterm birth), an information app (e-TOP) for parents was designed. The e-TOP app was developed in co-creation with parents, TOP interventionists, and experts in the field of prematurity with special knowledge of e.g. nutrition, sleep, long-term consequences of prematurity. Usability of this app will be evaluated separately and is outside the scope of this feasibility study.

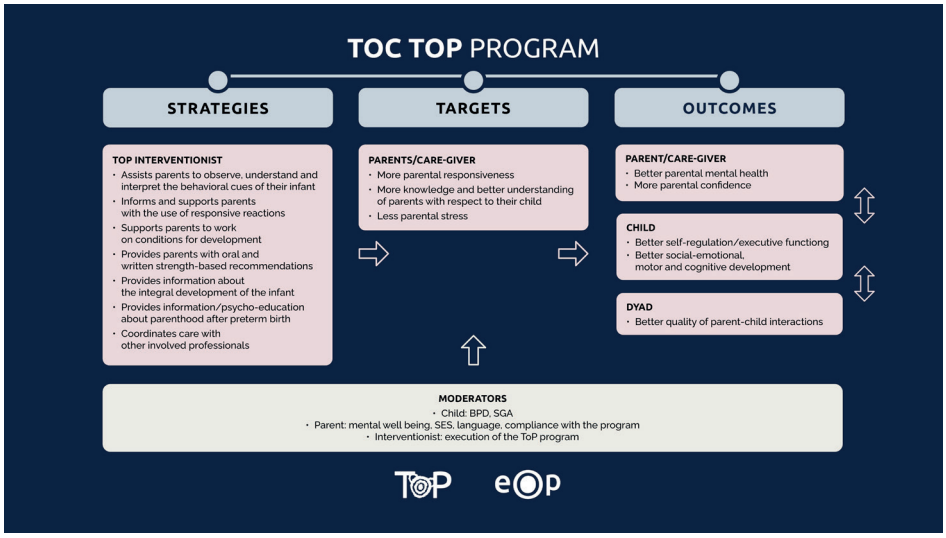

Figure 3.1. Theory of change of the TOP program.
Abbreviations: BPD, Bronchopulmonary dysplasia; SGA, Small for gestational age; SES, Socioeconomic status.

For this study, fifteen certified and experienced TOP interventionists received a 1-day training. The training was conducted by senior TOP-educators and members of the research team (MF, EM, MJ). The training contained substantive information about the target group, the use of the e-TOP app, the research protocol, including the measurements and Informed Consent procedure.

Participants

Infants were eligible to participate if (a) they were born with a GA between $32^{0/7}$–$34^{6/7}$ and a birth weight > 1500 g (as MP infants with a birth weight < 1500 g can already participate in the regular TOP program); (b) parents were able to understand and read the Dutch language or an interpreter could be arranged; (c) lived within acceptable traveling distance from a participating TOP-interventionist (approximately 20 min).

Infants were excluded if (a) the infant had congenital abnormalities; (b) participated in another parent-child intervention program. For multiple birth, only one infant was included in the study. A sample size of 40 infants was chosen to capture the social and medical heterogeneity in the study group, aiming at 15 families with a lower educational background defined as completed primary school, vocational education, lower or middle general secondary education.

Measures

Compliance and execution of the intervention (T1)

The TOP interventionist completed checklists after each home-visit, including registration of caregiver attendance during the home visit (fathers/mothers). The content of intervention-execution was assessed by the key strategies, questions, issues and topics that were addressed during each home visit.

Intervention acceptability (T1)

Parents completed a questionnaire containing questions about perceived appropriateness of the intervention and indicated how satisfied they were with the intervention on a scale of 1 (lowest) to 10 (highest). Following completion of the intervention, eight parents were interviewed by the first author (MF) to gain more in-depth information about their experience with the TOP program. Interviews (video or phone call) were offered at a time convenient for the parents. For the post-intervention interview we used the following five questions: 1) How is (name infant) doing? How are you, as parents, doing after the first months at home?; 2) Can you tell me how you experienced the TOP program and TOP interventionist?; 3) Could you tell if and how the intervention helped you with taking care of your preterm infant?; 4) Did you make use of other services or health care professionals during the intervention period?; and 5) Could you give us any suggestions or advice for improvements to the intervention?

Following completion of the study, interventionists were asked to participate in a focus group meeting to share their experiences with the execution of the intervention and participating in the study. The following themes and questions were discussed: 1) Overall thoughts about participating in the study and execution of the intervention; 2) Timing, frequency, duration of the TOP program for MP infants; 3) Used intervention tools (part of the intervention protocol); 4) Suitability of the TOP training to carry out the MP intervention; and 5) The use, feasibility, and content of the app. The evaluation of the fifth theme was not the focus of the current study and will be described separately.

Limited-efficacy testing

Child outcomes (T1)

Infant motor development was assessed at T1 with the Alberta Infant Motor Scale (AIMS) (28). The total score of the items of the four gross motor positions was used and translated to a percentile ranking compared to a normative age-matched sample of infants. A score below the 5th percentile, based on the Canadian norms, was used to define delayed motor functioning. The AIMS is a reliable and validated observational tool (29).

Parent report of infant social-emotional development at T1 was assessed with the Ages and Stage Questionnaire Socio-Emotional 6-month Dutch version, 2nd edition (SED: ASQ-SE2-NL). The parent-report screening instrument consists of 26 items related to developmentally appropriate behavior. Parents indicate whether the child shows the described behavior 'most of the time' (0 points), 'sometimes' (5 points), or 'rarely/never' (10 points). Additionally, 5 points were given if parents indicated being worried about the described behavior. Scores range from 0 to 145, with higher scores representing a worse socio-emotional development. The cut-off score of 30 points was used to define infants at risk for socio-emotional difficulties (30). The ASQ-SE-2-NL has good psychometric properties and clinical utility in screening for delay or problems (31, 32).

Parent outcomes (T0)

Parents completed the Distress Thermometer for Parents (DT-P) at T0. The DT-P measures overall distress using a thermometer score ranging from 0 to 10, with a score of 4 or higher indicating clinically elevated distress. It also evaluates everyday problems across six domains: practical (seven items), social (four items), emotional (nine items), physical (seven items), cognitive (two items),

and parenting (five items). Scores for each domain are calculated by summing the items marked as problems (yes = 1, no = 0). The DT-P is a widely used tool in clinical practice and has shown good psychometric properties (33, 34).

Parent outcomes (T0–T1)

Parenting self-efficacy was assessed at T0 and T1 with the Dutch version of the Maternal Self-efficacy in the Nurturing Role (SENR) (35). The total score of the 16 items, each rated on a 7-point Likert scale from 1 (Not at all representative) to 7 (Strongly representative) was used. Higher scores reflect greater feelings of competence in parenting. Previous studies have shown moderate to high test-retest reliability and good internal consistency (36, 37).

Parental reflective functioning with regard to their child was measured at T0 and T1 with the Parental Reflective Functioning Questionnaire (PRFQ). The PRFQ (18 items) assesses three domains: Certainty about mental states (ability to recognize the opacity of mental states), Interest and Curiosity in mental states (genuine parental interest in and curiosity about infant mental states), Pre-mentalizing (a parent's tendency to make negative attributions about their child's behavior) rated on a 7-point Likert scale ranging from disagree to agree. In this study all three domain scores were used. The PRFQ has good psychometric properties (38, 39).

Parental sensitivity was assessed with the parent scale of the Attachment During Stress scale (ADS) at T0 and T1 (40). The ADS can be used in mildly stressful situations, such as a doctor's visit, but also in daily situations such as bathing, dressing, and free play. The ADS can distinguish sensitive from less sensitive mothers. In this study, two structured parent-infant interaction tasks (diaper change and free play) were video-recorded by the TOP interventionists. For the diaper change, parents were instructed to undress their child, change the diaper, and then redress the child.

The undressing and dressing were filmed, but recording was paused during the actual diaper change for privacy. For free play, parents played or interacted with their child for 3 min, starting the recording after receiving instructions and stopping it after 3 min. Coding was conducted by two master students in pedagogy, trained by two researchers (EM, MJ). The ADS has seven parental sensitivity items (gazing, vocalizing, touching, holding, affect, proximity) rated on a 5-point scale, with scores of 1–2 suggest avoiding contact, 3–4 reflecting sensitive behavior, and 5 reflecting over-anxious interaction. A total score for parental sensitivity per task was calculated based on the frequencies of the

sensitive scores (scores 3 and 4) obtained for each of the behaviors displayed. The scores for parental holding were not used, as this behavior did not occur frequently enough. Thus, a score between 0 and 6 for parental sensitivity was possible, with 0 indicating the absence of any sensitive behavior, and 6 indicating all the behaviors displayed were sensitive. To determine interobserver reliability, 19% of the videos were double coded. Mean interobserver reliability (intraclass correlations; ICC) was 0.96 (range 0.93–1.00) for diaper change and 0.98 (0.89–1.00) for free play. The final scores for the double coded data were obtained by randomly selecting the scores of one of the observers.

Analysis

Quantitative data

Data were exported from the Castor database (Electronic Data Capture, Ciwit BV, Amsterdam, the Netherlands, 2021) to the Statistical Package for the Social Sciences version 28 (SPSS Inc. Chicago, Illinois). Baseline infant and parent characteristics, intervention data, child outcome measures (ASQ-SE-NL and AIMS) were summarized by descriptive statistics using the mean, standard deviation, frequencies, percentiles, or percentages.

The median and interquartile range (IQR) was used when the data were non-normally distributed. The change scores for the SENR and PRFQ were calculated at an individual level, using the Reliable Change Index (RCI). The RCI reflects the absolute change in scores pre- and post-intervention that are required to be confident that the observed change was not due to random variations over time. The ADS results were analyzed using Paired Sample t-tests. For the parametric tests, the effect sizes were calculated and interpreted according to the Cohens' d criteria (0.20 = small, 0.50 = medium and 0.80 = large) (41). Significance testing was not appropriate for this feasibility study due to the set-up and sample size (42).

Qualitative data

Qualitative data (audio recordings) from the interviews with parents and the focus group meeting with interventionists were evaluated by the first author (MF). This data was used to gain more insights about the usability and suitability of the intervention and to verify if findings correspond with the findings on the satisfaction questionnaire. Content related questions were used to identity points for improvement.

Results

Recruitment capability and compliance

Ultimately, ten interventionists were able to include MP families. The most common reasons for interventionists not including families were an unexpected smaller number of discharged MP infants during the inclusion period as reported by the hospitals. All of the collaborating hospitals were interested in the study and were in close contact with the TOP-interventionist.

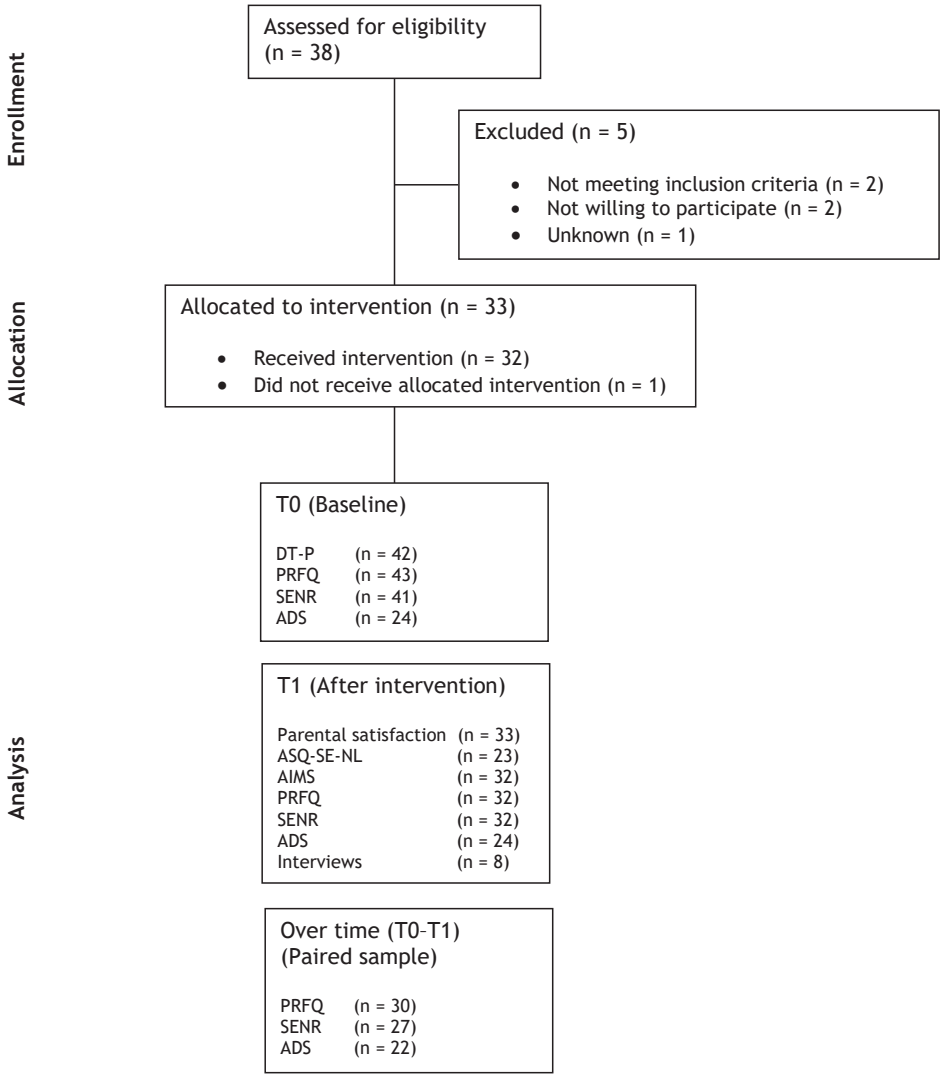

Figure 3.2. Flow of participants.

A total of 38 families were identified by hospital staff and were interested in the study. Thirty-three families were eligible and agreed to participate and gave informed consent. One family withdrew after signing IC, but before the first intervention home visit. Thirty families gave additional consent for filming the parent-child interaction (Figure 3.2).

The pre-intervention visit was on average 13 days after discharge, the first home visit (intervention) a month after discharge. All 32 families completed the intervention program and received the six home visits. Mothers were present almost all six home visits (80%). Fathers were present more than half of the home visits. Additional information about the actual execution of the intervention is provided in Table 3.1.

Table 3.1. Dosage and execution of intervention delivery

Parental attendance at home visits (range 0-6 home visits)	Mean (SD) or %
Mothers	
Attendance at home visits	5.75 (0.51)
Attendance of > 3 home visits	100%
Attendance of 6 home visits	79.9%
Fathers	
Attendance at home visits	2.56 (2.08)
Attendance of > 3 home visits	31.2%
Attendance of 6 home visits	12.5%
Topics *	**Mean (SD)**
Behavioral cues and responsive reaction	5.60 (0.81)
Sleep	5.16 (1.04)
Feeding	5.32 (1.35)
Motor development	5.51 (0.62)
Correcting for infant age	4.45 (1.68)
Transition to parenthood	3.29 (1.68)
Long term consequences of prematurity	1.51 (1.28)
Return to work	3.16 (1.71)
Health related questions	1.39 (1.22)
Follow-up/care after intervention	1.42 (1.52)

* Topics; Per home-visit (0-6) registered focus of treatment.

Perinatal and socio-demographic characteristics

Infant were born on average at a GA of 33.6 ± 0.7 weeks. Six children were small for gestational age. One infant was diagnosed with bronchopulmonary dysplasia. Within the study group, no severe complications such as necrotizing enterocolitis, IVH grade III/IV, PVL grade > 1 were reported in the medical history. Length of hospital stay (LOS) was almost 3 weeks, and infants were on average

discharged at $36^{5/7}$ weeks GA. Despite the efforts to include families with lower educational backgrounds, parents were mostly well educated, employed and had a family status of 2 parents. Parents reported an elevated level of distress at T0. The mean (SD) DT-P thermometer score was 3.68 (2.6) with the highest problem scores on the emotional, physical, and practical domains. Half of the parents scored within the clinical range of the DT-P (score ≥ 4). Mothers scored more often within the clinical range than fathers (62% vs 25%) (Table 3.2).

Intervention acceptability

Parents were very satisfied with the amount of home visits (6) and were very satisfied with the intervention (mean satisfaction rate of 9.4 on a scale of 0–10). Two parents indicated the need for prolongation of the intervention due to their infant's delayed development. In general, parents stated that the intervention was helpful to learn to understand the behavioral needs of their infant, they gained confidence in their caregiving and valued the strength-based parent report (Table 3.3).

Ten families were approached to participate in the interview after the intervention by first author (MF). Two families did not respond to the mail invitation and two families did not want to participate due to their busy schedules. Ultimately, six families participated, including six mothers and two fathers. All eight parents emphasized their need for appropriate support after discharge, they felt unprepared and insecure. All parents emphasized the importance of receiving support promptly, preferably within the first few days after discharge. They expressed that the intervention fitted their needs and described it as most suitable care. Participating in the intervention provided them with the care they needed, and they hoped this intervention would become available for other parents. Parents valued the interventionists for their expertise and knowledge on the impact of prematurity, their accessibility, and practical guidance. Parents indicated again that the amount of six home visits was sufficient and valued the individualized parent report after the home visit. They assigned their enhanced self-efficacy and understanding of their infant's needs to the intervention (Table 3.3).

Ten TOP interventionists evaluated the MP intervention during a focus group meeting. They all agreed that in general six home visits were sufficient but also indicated the need for flexibility to schedule additional visits if necessary for the infant or family. They emphasized the need for a rapid start of the intervention, preferably within the first week after discharge. They reported that the implementation of the TOP program strategies within this target group was well

applicable and that their extensive expertise with VP infants was easily transferable to the new target group. They also indicated that specifically parents with a lower educational background found the Informed Consent procedure, videotaping during the pre-home visit and the extensive data collection somewhat threatening.

Table 3.2: Baseline clinical and demographic characteristics of the families

Infants		At discharge	
Gestational age (GA), week, mean (SD)	33.6 (0.7)	Length of stay (LOS), days, median (IQR: Q1–Q3)	19.5 (15.3–28.5)
Birth weight (BW), g, mean (SD)	2201 (379)	Postmenstrual age at d/c, days, mean (SD)	257.3 (7.5)
Small for gestational age (SGA), n (%)	6 (19)	Time between d/c-1 pre-visit (days), median (Q1–Q3)	13.0 (9.0–23.3)
Sex, male/female, n (%)	17/15 (53%/47%)	Time between d/c-1 intervention, median (days), median (Q1–Q3)	31.0 (24.3–48.8)
Number of twins n (%)	5 (15.6%)		
Apgar score at 5 min, mean (SD)	8.5 (0.3)		
Bronchopulmonary dysplasia, n (%)	1 (3.1%)		
Necrotizing entercolitis, n (%)	0 (0%)		
IVH grade III/IV, n (%)	0 (0%)		
PVL grade > 1, n (%)	0 (0%)		
Parental characteristics	**Mothers (n=29)**	**Fathers (n=14)**	
Age, years, mean (SD)	30.9 (4.3)	33.4 (4.8)	
Firstborn child, n (%)	19 (65.5%)	7 (50%)	
Family status of 2 parents, n (%)	32 (100)		
Country of birth, n (%)			
Netherlands	28 (96.6)	14(100)	
Turkey	1 (3.4)		
Dutch language spoken at home, n (%)	43 (100)		
Education, n (%)			
Lower	1 (3.4)	1 (7.1)	
Intermediate	13 (44.8)	5 (35.7)	
High	15 (51.7)	8 (57.2)	
Job			
Fulltime	15 (51.7)	12 (85.7)	
Parttime	14 (48.3)	2 (14.3)	

SGA was defined as < P10 with the reference data from https://www.perined.nl/onderwerpen/geboortegewichtcurven BPD was defined as Oxygen dependent after GA 36 weeks NEC was defined as stage ≥ according to Bell's clinical staging Periventricular Leukomalacia (PVL) was defined when ≥ 1 Intraventricular hemorrhage (IVH) was defined when > 3. Highest educational level completed: Low level education refers to primary school, vocational education, lower of middle general secondary education. Intermediate: refers to higher secondary general education, pre-university education. High; Higher vocational or university. This classification is based on the 'Standaard Onderwijsindeling 2021' (25).

Table 3.3. Satisfaction with the TOP program

Questions	Mean (range) or yes (n) no (n)	Comments parents
How would you rate the intervention you received (1-10) (n = 33)	9.39 (8-10)	- *The TOP program was very valuable for us.* - *The best guidance we received.* - *Home visits were wonderful.*
1. Was the amount of home-visits sufficient?	Yes (31) No (2)	- *I am still insecure about my baby's development, would have liked more home visits.* - *Six months is a good period for the intervention. I do feel that everything is going as it should be right now.*
2. Did the intervention help to understand behavior and needs of my child	Yes (32) No (1)	- *I do not think I would have become such a sensitive parent without the guidance in how to read my baby's signals.* - *I do know better when he's ready to make contact and when he needs some rest.*
3. Did the intervention help to gain confidence in caregiving	Yes (29) No (1) Unknown (3)	- *My insecurities about does he have pain, can he be on his tummy, what is good for him would be discussed during the home visits.* - *She helped me understand and gave support and trust in myself.*
4. Did the conversation card help to formulate your questions and concerns	Yes (8) No (2) Not used (23)	- *It helped me to formulate my concerns and prepare the home visit.* - *Did not use it, use my phone to note my questions.* - *Gives structure and helps me to keep overview.*
5. Did you find the parent report of additional value	Yes (32) No (1)	- *Good to receive specific information about my child and be able to see the progression.* - *Give ability to read all the information given in the home visit.* - *Pictures give insights and showed my babies change.*

Limited efficacy testing

Child outcomes

For motor development, infants' mean AIMS score at T1 was 24.8 (SD 6.1). Three infants (9%) scored below the cut-off score of P5. Regarding socio-emotional development, the median score on the ASQ-SE2-NL was 20.0 (IQR 13.8, 26.3). Two infants (6%) were identified as at risk for delay (Table 3.4).

Table 3.4. Descriptive statistics and effect sizes for infant and parent outcome measures

Measure	T0	n	T1	n	Percentage above/ below cut-off *	n	
AIMS raw score, M (SD)			24.1 (5.5)	32	9.4		
Prone			9.7 (3.1)				
Supine			8.2 (1.3)				
Sitting			4.9 (2.2)				
Standing			2.1 (0.82)				
ASQ-SE-NL, median (IQR: Q1-Q3)			20.0 (13.8-26.30)	32	6.7		
	T0 M (SD)	n	T1 M (SD)	n	Cohen's d	n	Improvement T0-T1 (no., %) ***
SENR	90.60 (11.9)	41	94.80 (9.7)	32	.35	27	7/27 (26)
PRFQ-PM **	1.64 (0.64)	43	1.55 (0.52)	32	.59	30	9/30 (30)
PRFQ-CMS	4.11 (0.84)	**43**	4.55 (0.77)	**32**	.81	30	12/30 (40)
PRFQ-IC	5.34 (0.91)	43	5.39 (0.89)	32	.56	30	1/30 (3)
ADS		24		24		22	
Diaper change	5.64 (0.70)		5.88 (0.33)		.40		
Free play	5.17 (1.2)		5.63 (0.57)		.38		

* Used cut-off score: AIMS < P5, ASQ-SE2-NL > 30 pt.
** PRFQ-PM refers to domain: Pre-mentalizing, PRFQ-CMS refers to domain Certainty about mental States PRFQ-IC refers to domain Interest Mental states.
*** Used RCI index: SENR 10.57, PRFQ-PM 0.81, PRFQ-CMS 1.19, PRFQ-IC 0.97.

Parental outcomes

Parental sensitivity, as measured with the ADS at T0, was high for both tasks, diaper change (M 5.64, SD 0.70) and free play (M 5.17, SD 1.20) and improved over time (Cohens' d 0.40 and 0.38 respectively).

Parents' perception of their competence in caring for their infant as measured with the SENR showed a high initial score at T0 (M 90.6, SD 11.9) and increased 4.2 points (M 94.8, SD 9.7). The effect size was small (Cohens' d = 0.35) and 26% of the parents reported a clinically relevant improvement in their confidence in parenting skills (RCI > 10.57). Parents' reflective functioning as measured with the PRFQ (three dimensions) improved over time. Certainty about mental states improved from T0-T1 with a large effect size (Cohens' d 0.81), 40% of the parents showed a clinically relevant improvement (RCI > 1.19). For the domains Interest and Curiosity and Pre-mentalizing medium effect sizes were found (Cohens' d 0.56 versus 0.59). The RCI indices for the PRFQ interest and

Curiosity, showed only for 1 participant a clinically relevant positive change (RCI > 0.97). For the PRFQ domain pre-mentalizing, 30% of the participants showed clinical important progression (RCI > 0.81) (Table 3.4).

Discussion

The ability to enroll and retain families and the positive results of this study fully support the feasibility of the TOP program for the new target population of MP infants and their parents. The three feasibility dimensions; recruitment capability/compliance, acceptability, and limited efficacy testing will be discussed separately.

Recruitment capability and compliance

The willingness of the clinical staff at the participating hospitals to inform parents, and parents wanting to participate in this intervention confirmed the observed need for support after discharge in studies from Davis-Strauss (43) and Adama (44). Taking into consideration that the TOP program is offered preventively, our enrollment feasibility rate and the compliance with the intervention was remarkably high compared to other parenting intervention studies (45, 46).

Non-participating families (6) of 38 interested families wanted to receive the TOP program, however participating in the study was too much to ask. They found the Informed Consent letter difficult to understand, and despite the fact the video recording of the ADS before the intervention was not mandatory, parents perceived it as an assessment for their parenting skills. These unintended consequences of participating in a study could have led to less already disadvantaged participants. A postponed informed consent procedure, availability of translated documents, support to complete questionnaires, and text to speech features might be useful to explore in future intervention research.

For feasibility research, findings about compliance with the data collection is also important. Via questionnaires, parents cited reasons for not completing assessments at T0 and/or T1, including returning to work, feeling overwhelmed by life stressors such as moving, and the amount of self-reported measures for both fathers and mothers. These findings advocate for brief self-reported measures to reduce respondent burden and facilitate involvement of fathers.

Intervention acceptability

All families adhered to and engaged in the intervention. De-briefing post-intervention interviews showed acceptability and suitability of the strategies on improving parent-child interaction. Another important aspect parents appreciated was the collaborative relationship with the TOP-interventionist. The importance of promoting parent-infant interaction and trust between parent and the interventionist for successful participation, is confirmed by Oberg in a qualitative systematic synthesis (21).

The timing of the first intervention session, in our study one month after discharge, requires adjustment. Both parents and TOP interventionists expressed the need for support to start earlier, ideally within the first two weeks post-discharge. Starting the TOP intervention immediately after hospital discharge would assist parents in managing their insecurities and enhancing practical skills during the transition from hospital to home. In this feasibility study, informed consent and reflection time to consider participation delayed the first home visit. Including families already during hospital admission may decrease this time.

Limited efficacy testing

Child outcomes

The post-intervention level of motor development at 6 months CA surpassed our expectations (Mean AIMS score of 24.1). Recently, Suir et al. found lower mean scores (18.5–22.4) in a general Dutch population than in our sample (47). The execution of the responsive strategies containing holding, positioning, and stimulating mobility activities for the infants could have improved the motor development in our sample, especially in the age-related supine and prone position. To learn if the positive motor development persists with achieving new motor milestones, we would recommend monitoring the gross motor trajectory over a longer period. After all, research showed low predictive values of the AIMS (4 months: 40%; 8 months: 66%) due to intra-individual variability (48, 49).

Another consideration regarding the high AIMS scores is that TOP interventionists, who are experienced pediatric physical therapists skilled in scoring motor development using the AIMS, were directly involved in the intervention, and not blinded. For future studies, we recommend video-recording the motor development assessment, allowing independent, blinded researchers to conduct the scoring.

Parents reported a positive socio-emotional development of their MP infants. All aspects of the measured socio-emotional development (i.e., behavior regulation, feeding, sleeping, and interaction) were addressed during the home visits, in the customized parent reports, and additional information was provided via the e-TOP app. These deployed intervention strategies could have contributed to the positive impact on the parent-reported socio-emotional development of their child. Although the ASQ-SE2-NL was useful to open the conversation with parents as a guidance for intervention strategies, we need to be careful to draw conclusions based on these outcomes. Low sensitivity indices and low predictive value at the age of 6 months was reported by de Wolff (31) and Krijnen (32) and we therefore recommend longer follow-up to evaluate if these short-term results perpetuate.

Parent outcomes

We found improvements on all parental measures (SENR, PRFQ, ADS) with small to large effect sizes. We speculate that the intervention strategies have contributed to the positive outcomes in parents' feelings of self-efficacy, reflective functioning, and parental sensitivity. During the home visits, the primary focus was on observing and understanding the infant's behavioral cues across various developmental domains, such as interaction, sleeping, and feeding. Subsequently, the appropriate parental responsive reaction, to meet the infant's emotional and developmental needs, were identified and practiced with the parents. Although our results were very promising, the effect of time and assessing efficacy of the intervention on the parental outcomes cannot be determined with the single group design and would require a control group.

The pre-intervention scores on parental self-efficacy were unexpectedly high compared to the baseline high distress scores on corresponding DT-P domains 1) emotional problems containing questions about self-confidence, feelings of depression, and 2) parenting problems in caring, feeding, and development of the child. Our findings subscribe the view of de Moor that in addition to measuring distress, assessing parenting self-efficacy during an intervention can help to understand parents' appraisal of one's ability to parent (37).

This feasibility study confirmed the need for post-discharge intervention for MP infants and the TOP program positively affected the infants and their parents. By pairing our surveys and checklists with qualitative data we gained valuable information about suitability and the impact of the used TOP-strategies. The use of qualitative measures (interviews, ability to comment in the questionnaires,

focus group) added to the understanding of modifiable factors influencing the recruitment outcomes, and compliance with data collection.

Limitations

The present study is the first step to explore if moderate preterm born infants and their parents benefit from the modified TOP intervention. There are several limitations to the present study.

This feasibility study was conducted with a small sample size and was not set up to compare between groups, limiting our analyses of potential efficacy. We need to advance to a larger, randomized controlled trial design to be able to confirm the positive study results, and to be able to better identify mechanisms of change and interrelationship between outcomes. The used measures relied partially on subjective self-report measures, which may be biased by social desirability. The use of direct measures, such as the ADS, AIMS and the qualitative data however endorse the found positive findings.

Both fathers and mothers were invited to participate in the study and all signed informed consent. Fathers participated on average in 2.5 (out of six) home-visits, and only a limited number of fathers completed the questionnaires, and we could only compare the pre-post data of seven fathers, which is insufficient to draw conclusions. Dutch fathers have limited parental leave and were less present during the home visits which may have also impacted the compliance with the data-collection. We would recommend more flexibility in work-schedules of the interventionists to promote paternal attendance.

Although the level of parents' education differed, almost all participants received higher secondary general education and were all employed. The inclusion criteria such as language, the strict research protocol including the informed consent procedure, and the number of digital questionnaires might have led to underrepresentation of families with different backgrounds and lower educational levels. During the initial phase of developing the MP intervention and the e-TOP information app, partnership with low literacy end-users has been successful. To make studies and implementation trials more accessible and meaningful for participants, we would recommend involving parents or parent representatives throughout the entire research cycle, including design and choice of measures (50).

Additional findings from the interviews and a focus group contained valuable information and were very much in favor of the feasibility of the intervention. It confirmed the high parental satisfactory rates and need for support. However,

the re-listening of the audiotapes, summarizing the transcripts and the evaluation of the focus group results were all executed by the first author and discussed with the research team. To limit biases in data analyses, we recommend having double coders for the qualitative data in future research.

Conclusion

The Dutch post-discharge parenting intervention (TOP program) is feasible to use with moderate preterm born children and their parents. The findings of this study indicate that the modified TOP program meets the needs of MP infants and their parents, resulting in a high intervention adherence and acceptability, promising child outcomes and positive changes in caregiver sensitiveness, self-efficacy, and reflective functioning. This study contributes to the preparatory work prior to a larger scale evaluation and dissemination.

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Chapter 4

Feasibility of a preventive parenting intervention for very preterm children at 18 months corrected age: A randomized pilot trial

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Abstract

Objective: To evaluate the feasibility and potential efficacy of an age-appropriate additional parenting intervention for very preterm born toddlers.

Study design: In a randomized controlled pilot study, 60 of 94 eligible very preterm born children who had received a responsive parenting intervention in their first year were randomized to usual care or the additional intervention, consisting of 4–6 home visits between 18 and 22 months corrected gestational age (CA). Parents were supported to responsively interact during increasingly complex daily activities and play. Parental satisfaction with the intervention was evaluated with a questionnaire. At baseline and 24 months CA, parents completed the Infant Toddler Social and Emotional Assessment, the Ages and Stages Questionnaire, and the Dutch Schlichting Lexilist for receptive language. At 24 months CA, motor, and cognitive development was measured by the Bayley Scales of Infant and Toddler Development, Third Edition Dutch version, and parent-child interaction was evaluated by the Emotional Availability Scales.

Results: Parental compliance and satisfaction with the intervention was high. Effect sizes (after correction for baseline variables) were small for internalizing and competence behavior, receptive language, and problem solving; medium for cognitive development and parent-child interaction; and large for externalizing and dysregulation behavior and motor development.

Conclusion: After a post discharge intervention during the first year, an additional responsive parenting support at toddler-age is feasible and associated with positive outcomes in a broad array of parental and child outcome measures.

Introduction

Very preterm birth (< 32 weeks of gestation) and very low birth weight (< 1500 g) are strongly associated with developmental and behavioral problems (1-4). In response, various post discharge intervention programs have been developed. The magnitude of the effects, however, has been modest, and the challenge for future programs is to design and test resources and intervention strategies that improve the preterm child's development and participation later in life (5-8).

The Infant Behavioral Assessment and Intervention Program (IBAIP) is a post discharge preventive neurobehavioral intervention programs that yields long-term benefits for the very preterm child's development (9). The program consists of 6–8 home visits from discharge until 6 months corrected gestational age (CA) and has been evaluated extensively with follow-up studies until 5.5 years' CA in the Netherlands. Better cognitive, motor, and behavioral outcomes were found at 6 months' CA, and better motor outcome at 12- and 24-months' CA (10, 11). At 5.5 years' CA, the intervention group had better visual-motor outcomes, better verbal IQ, and lower frequency of a performance IQ less than 85 (12, 13).

Aiming to further boost the results of the IBAIP intervention, an early intervention program providing transmural developmental support for preterm infants and their parents (TOP program) was developed and implemented as a standard of care in the Netherlands in 2010. The TOP program is available for children born with a gestational age < 32 weeks and/or birth weight < 1500 g. It consists of 12 home visits from discharge until 12 months (CA) and is carried out by specially trained pediatric physical therapists. The focus of the TOP program is to strengthen the parent's well-being and sensitive responsive parenting, to enhance the infant's self-regulatory competence and exploratory participation, and to diminish stress, because these aspects play a central role for a favorable development of young infant's brain (5, 14).

Recent literature has emphasized the importance of consistent parental responsiveness across both infancy and the toddler preschool period for behavioral and cognitive development in term infants and even more in preterm infant (15, 16). In addition, if the intervention coincides with the sensitive periods for these developmental domains they might improve optimally (17-20). If so, this may require parental understanding of children's changing developmental needs over time. Consequently, we wanted to evaluate a staged curriculum that begins with the TOP intervention during the first year of life, followed by

an additive responsive parenting program (the TOP+ program) during the age of 18–22 months.

The primary aim of this pilot study was to examine the feasibility of the additional TOP+ intervention. In addition, we wanted to explore whether the additional intervention was favorable for cognitive, motor, and behavioral development of the child and the parent-child interaction.

Methods

Families were eligible for the study if they had received the TOP program during the first year; the child had not been diagnosed previously with cerebral palsy, Down syndrome, or participated in another intervention program; the parents were sufficiently able to understand the Dutch or English language; and the child lived within a reasonable travel distance from the therapist's working area.

For the purpose of the TOP+ program, 5 TOP physical therapists received additional training on age-specific aspects of behavioral expressions, self-regulation, and the different developmental domains, with a special focus on preverbal communication, play, and the use of scaffolding techniques. The TOP+ program involved a minimum of 4 in-home sessions (60–90 minutes each session) when the infant was between the age of 18- and 22-months' CA. In addition, 1 or 2 in-home sessions could be used if the physical therapists felt that more contact to optimize the intervention was needed.

The TOP+ program uses a process and strength-based approach to support well-tuned and matched parent child interactions. Parents are encouraged to follow their child's interest to positively engage in daily activities, such as eating and dressing, and age-specific activities with their child, including free play or shared book reading. The therapist provides the parent with short comments about the child's expressions of positive engagement, self-regulatory strategies, or efforts. Accordingly, the therapist may give suggestions how the parent can give information in a scaffolding way (physical or verbal support, or by structuring the ask), or how the parent can co-regulate and support the child's feeling of comfort and safety. After each session, a written report is made for the parents, including strength-based recommendations with supporting photos. The same therapist who delivered the TOP program and who was familiar to the family was assigned, when possible.

To maintain the consistency of the intervention, written parent reports were regularly reviewed and supervised by the trainer. Monthly meetings with

the 5 therapists were scheduled to share experiences and discuss aspects of the intervention.

Eligible families received an information folder and invitation to participate in the study. After written informed consent was obtained by the TOP physical therapists, the children were assigned randomly to the TOP+ intervention group or the control group. A computer-generated block randomization was performed, stratified for therapist. Children from multiple births were assigned to the same group because of the nature of the intervention. Perinatal variables were abstracted from medical records. Socioeconomic data were obtained by parent interview. Three questionnaires were completed by the parents at baseline (18 months' CA) and at the follow-up assessment (24 months' CA). At that time, the Bayley Scales of Infant and Toddler Development, Third Edition, Dutch Version (BSID-III-NL) (21) was conducted by independent specialized psychologists blinded to group assignment. The BSID-IIINL is part of the standard follow-up protocol at 24 months and was conducted in either the Academic Medical Center or Free University Medical Center in Amsterdam.

A videorecording to evaluate the parent-child interaction with the Emotional Availability Scales (EAS, Fourth Edition) (22) at 24 months CA was done by a researcher in a free play situation at the home of the children. Scoring was done by an EAS-certified psychologist blinded to group assignment. Parents who received the additional TOP+ intervention filled out a questionnaire to evaluate their satisfaction with the intervention. Because this was a pilot study, no sample size calculations were performed; however, we aimed to include 30 children per group to capture the diversity of this group. The study was approved by the Medical Ethics Committee of the AMC, the Netherlands.

The Infant Toddler Social and Emotional Assessment (ITSEA) is a parent-report questionnaire developed to assess social-emotional problems and competencies in 12- to 36-month-old children. It consists of 166 items that measure 17 syndrome scales and 4 domains via a 3-point Likert rating scale (0 = not true/rarely, 1 = somewhat true/sometimes, 2 = very true/often, "No opportunity" available for some items). Based on these ratings, age- and sex-specific t scores (mean = 50, SD = 10, range 25–80) were calculated for the 4 domains: social-emotional competence; externalizing behavior problems; internalizing behavior problems; and dysregulation. The ITSEA has good test-retest reliability, good criterion validity, and a well-supported factor structure (23).

The Ages and Stages Questionnaire, Third Edition (ASQ-3) is a screening questionnaire to detect developmental delays in children. For this study, we used

the ASQ-3 at 18 and 24 months. The questionnaire consists of 30 developmental items to assess 5 domains of child development: communication, gross motor, fine motor, problem solving, and personal social. For each item, the parents indicate “yes” (10 points), “sometimes” (5 points), or “not yet” (0 points) to represent their child’s ability to perform a task. A greater score indicates better development. Scores for each domain and an overall score can be calculated. The presence of any domain < 2 SD below the mean was considered a positive screen for “at risk” for developmental delay. Because there was no standardization for Dutch children at the time of the study, normative values from the US were used. Reliability and specificity are high, and sensitivity ranges from 51% to 90% (24).

The Dutch Schlichting Lexi-list for receptive language was used at both 18- and 24-months’ CA (25). This parent-reported list consists of 225 words and small sentences that children between 15 and 25 months can understand. Dutch age norms for each month are available. The test score was converted into a standard score (Lexi-list Perceptive language quotient), with age norms for each month (mean = 100, SD = 15). The Schlichting Lexi-list is a valid and reliable measure (25). Parents who were included in the TOP+ program evaluated the usefulness and duration of the intervention with a questionnaire that addressed 4 aspects (practicability, knowledge transfer, collaboration, and amount of sessions) of the intervention. The questionnaire consisted of 13 multiple-choice, 3 open questions, and 1 question to grade the usefulness of the intervention.

The BSID-III-NL and US norms were used to assess the cognitive and motor development at the CA of 24 months (21, 26). The BSID-III-NL is normative, value-referenced assessment. The composite scores for the cognitive and motor scales are age standardized with means of 100 and SDs of 15. The Fine Motor and Gross Motor subscales scores are age standardized with a mean (SD) score of 10. The BSID-IIINL was found to be a reliable and valid instrument with good psychometric characteristics for Dutch children and high reliability on the subtests.

The EAS is a measure of the overall quality of the observed emotional availability in parent-child interactions. The EAS consists of 6 dimensions: 4 dimensions of parental behavior (sensitivity, structuring, non-intrusiveness, and non-hostility) and 2 dimensions of child behavior (responsiveness and involvement) (27, 28).

Statistical analyses

Data were analyzed with the Statistical Package for the Social Sciences version 20.0. (SPSSInc, Chicago, Illinois). Differences in sociodemographic and perinatal characteristics were analyzed with the independent-samples t test or c^2 tests. ANOVA was used to analyze group differences on the ITSEA, Lexi list, and ASQ-3 at 18 and 24 months and the BSID-III-NL and EAS at 24 months. In addition, ANOVA corrected for pre randomization baseline differences (gestational age) and known influential factors (small-for-gestational age, sex, maternal education) was performed for the BSID-III-NL and EAS. Effect sizes were expressed in Cohen d, which is interpreted as follows, > 0.2 small, > 0.5 medium, and > 0.8 large effect (29). The domain and total scores of the ASQ-3 at 24 months CA were analyzed with ANOVA and the frequency of "at risk" for developmental delay with logistic regression analyses, both performed with the ASQ-3 at 18 months as covariate. For the ITSEA and Lexi-list, repeated-measures ANOVA was performed with and without covariates.

Results

Enrollment for the study began in March 2013 and the study was completed in August 2014. Ninety-four very preterm infants who completed the TOP intervention and reached the adjusted age of 18 months were assessed for eligibility. Fourteen families could not be traced, and 20 families decided not to participate. After informed consent, infants were assigned randomly to the intervention group (n = 30) or control group (n = 30) (Figure 4.1).

The nonparticipating group did not differ from the participating group in gestational age (p = .23) and birth weight (p = .60) or length of stay at the hospital (p = .78). Mothers of the participating group had a significantly greater mean age (32 years) compared with the nonparticipating mothers (30 years) (p = .04).

All 30 infants of the intervention group received the planned 4–6 home sessions and written reports; 22 children (73%) were seen by their familiar TOP physical therapist. Twenty (80%) caregivers filled in the evaluation form after the intervention was completed. For twins, only 1 evaluation form was provided. Overall, parents were very satisfied with the intervention. All parents (100%) evaluated the number of sessions as adequate, the interventionist knowledgeable (95%), and the recommendations of the interventionist's valuable (95%). Parents gave the additional intervention a mean satisfaction rating 8.9 on a scale of 0–10, with a greater score indicating more satisfaction.

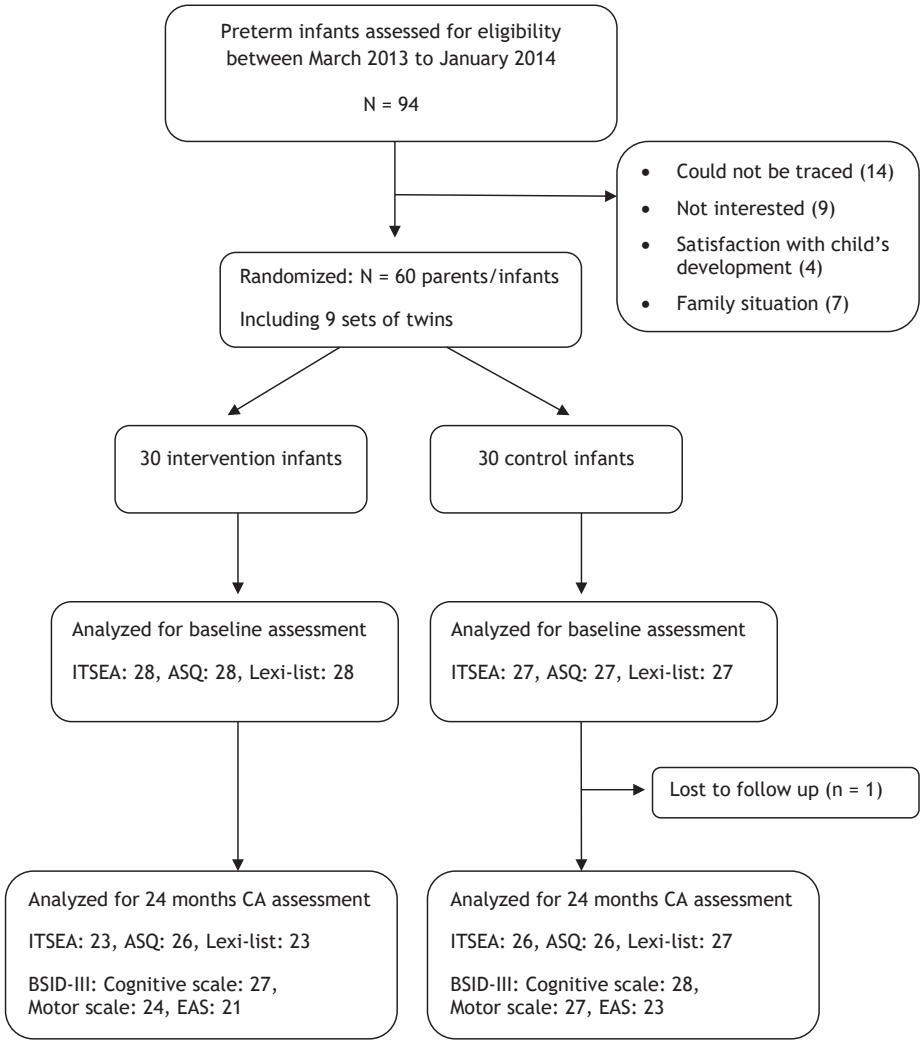

Figure 4.1. Flow chart of participants.

Despite randomization, infants in the intervention group had a lower mean gestational age than children in the control group (29.0 vs 30.9 weeks, p = .01). In addition, the infants in the intervention group had greater rates of ventilation support (p = .02) and oxygen support > 28 days (p = .03). The intervention group had fewer male subjects and small for gestational age infants than the control group, but these differences were not significant (Table 4.1).

Table 4.1. Perinatal and social background characteristics

Characteristics	Intervention group n = 30	Control group n = 30	p-value
Perinatal factors			
GA, weeks, mean (SD)	29.0 (17.5)	30.9 (17.9)	.007*
GA < 28 weeks (%)	10 (33)	6 (20)	.24
Birth weight, g, mean (SD)	1182 (430)	1353 (372)	.10
SGA (%)	7 (23)	13 (43)	.20
Sex, male/female	12/18 (40%/60%)	18/12 (60%/40%)	.12
Twins (%)	10 (33)	6 (20)	.24
APGAR score at 5 min, mean (SD)	7.7 (1.5)	7.7 (1.7)	.93
Ventilation (%)	19 (63)	10 (33)	.02*
Days of ventilation, mean (SD)	3.1 (7.9)	1.7 (3.9)	.39
Oxygen therapy > 28 days (%)	15 (50)	7 (23)	.03*
Oxygen therapy > GA 36 weeks (%)	7(23)	3 (10)	.17
Surfactant use (%)	15 (50)	8 (27)	.06
Dexamethason use (%)	2 (7)	2 (7)	1.0
Indocid/Ibuprofen use (%)	4 (13)	3 (10)	.70
Necrotizing enterocolitis (%)	1 (3.3)	0 (0.0)	.31
Septic periods before discharge (%)	16 (53)	10 (33)	.12
IVH grade I+II/III-IV (%)	4/1 (13/3)	3/0 (10/0)	.54
PVL	0	0	
At discharge			
LOS, days, mean (SD)	65.0 (29.8)	51.8 (32.7)	.11
Post menstrual age at d/c, wk, mean (SD)	38.3 (2.6)	38.0 (2.9)	.97
Breast milk at discharge (%)	22 (73)	20 (69)	.71
Oxygen supply at discharge (%)	0 (0)	2 (7)	.15
Monitor at discharge (%)	0 (0)	1 (3.3)	.31
Social factors			
Firstborn child (%)	12 (40)	16 (53)	.30
Family status of 2 parents (%)	28 (93)	27 (90)	.64
Mother born in the Netherlands (%)	23 (77)	22 (73)	.76
Father born in the Netherlands (%), n = 57	22 (76)	15 (54)	.08
Maternal education (%)#			
Low	3 (10)	3 (10)	.60
Middle	7 (23)	4 (13)	
High	20 (67)	23 (77)	
Paternal education (%), n = 57			
Low	6 (21)	6 (21)	.99
Middle	7 (24)	7 (25)	
High	16 (55)	15 (54)	
Language spoken at home (%)			
Dutch language	22 (73)	19 (63)	.39
Dutch and other language	4 (13)	5 (17)	
Exclusively other language	4 (13)	6 (20)	

Abbreviations: d/c, discharge; GA, gestational age; IVH, intraventricular hemorrhage; LOS, length of stay; PVL, periventricular leukomalacia; SGA, small for gestational age. # Low level education refers to primary school, pre- vocational/secundary school (< 12 years), middle educational level refers to senior general education, pre university education of secondary vocational education (13-16 years), high educational level refers to higher professional education of university (> 16 years).

Table 4.2: Comparison of the outcomes of the intervention group and control group on ITSEA, ASQ-3 and Lexi list

Outcome	18 mo Intervention mean (SD)	18 mo Control mean (SD)	24 mo Intervention mean (SD)	24 mo Control mean (SD)	p-value	p-value adjusted [d]	Cohen's d [c] (95% CI)
ITSEA							
Internalizing [a]	45.3 (12.0)	43.5 (7.6)	44.6 (12.5)	44.5 (8.9)	.54	.24	0.35 (-0.22; 0.92)
Externalizing [a]	47.3 (6.1)	46.0 (11.3)	44.2 (6.4)	48.8 (9.7)	.00*	.01	0.91 (0.26; 1.56)
Dysregulation [a]	50.0 (10.3)	48.0 (10.7)	47.3 (10.9)	50.0 (8.7)	.01	.05	0.63 (0.04; 1.22)
Competence [b]	50.6 (11.3)	50.1 (10.6)	53.2 (9.2)	50.3 (10.7)	.22	.59	0.17 (-39; 0.73)
ASQ							
Communication	32.4 (14.1)	25.5 (11.0)	50.6 (13.3)	44.6 (14.0)	.72	.63	0.14 (0.42; 0.70)
Gross Motor	47.7 (13.8)	46.4 (16.4)	48.9 (14.3)	48.9 (9.4)	.87	.39	0.29 (0.27; 0.85)
Fine Motor	50.9 (6.9)	47.9 (9.7)	50.7 (6.3)	50.4 (7.0)	.78	.38	0.29 (0.27; 0.85)
Problem Solving	40.6 (14.5)	37.9 (13.2)	47.7 (7.6)	45.8 (9.0)	.51	.38	0.29 (0.29; 0.87)
Personal Social	40.5 (8.2)	43.9 (7.9)	49.1 (8.9)	43.6 (9.9)	.03*	.04	0.67 (0.09; 1.25)
ASQ total score	179.6 (28.1)	176.0 (25.0)	247.0 (35.5)	233.6 (35.1)	.42	.59	0.17 (0.39; 0.73)
ASQ < -2SD, n (%)	10 (36)	8 (31)	2 (9)	5 (20)	.63	.46	NA
Lexi-list							
LBQ	96.2 (15.6)	92.1 (16.7)	99.6 (19.3)	90.4 (21.7)	.45	.22	0.35 (0.23; 0.93)

Abbreviations: LBQ, Lexi-list Perceptive language quotient; N/A, not applicable.
[a] Higher scores indicate more problem behavior.
[b] Lower scores indicate less optimal behavior.
[c] > 0.2 small effect, > 0.5 medium effect, > 0.8 large effect.
[d] Adjusted for gestational age, Small for GA, gender and maternal education.

There were no significant differences in scores at baseline (18 months' CA) between the intervention and control group on the ITSEA, Lexi-list, and the ASQ-3 developmental domains and total score. The control group scored slightly greater on the ASQ-3 domain communication (p = .05).

At 24 months, we observed a significant improvement over time in the intervention group on the ITSEA for externalizing behavior (p = .00) and dysregulation behavior (p = .01). After adjustment for baseline variables, the effect size was large for externalizing behavior and medium for dysregulation behavior. On the Lexi-list, a nonsignificant effect for the time by group interaction was found (p = .22; Table 4.2). At 24 months, the control group had a mean Lexi-list Perceptive language quotient of 90.4, indicating a small delay in receptive language. Only small, nonsignificant effects were observed on the ASQ domains between the intervention and control group at 24 months. On the personal-social domain, a significant difference was found in favor of the intervention group (p = .03). This difference remained significant after adjustment for gestational age, small for gestational age, maternal education, and sex (p = .04) with a medium effect size (Table 4.2).

At 24 months CA, the intervention group had a significantly greater score on the motor composite score (p = .04). After adjustment, the scores for both the cognitive (p = .02) and motor domain (p = .01) were significantly greater in the intervention group (Table 4.3). Effect sizes were large for the motor domain and gross motor subtest. A medium effect size was found for the cognitive domain.

There were no significant differences between the groups on the 6 domains of the EAS measured at 24 months CA. After adjustment, the parents in the intervention group had significantly greater (better) scores on 2 maternal scales (sensitivity [p = .01] and structuring [p = .02]), and on 1 dimension of child behavior (child involvement [p = .02]). Medium-to-large effect sizes were found on the different EAS domains, except for non-hostility (small effect) (Table 4.3).

Table 4.3. Outcomes on the BSID -III-NL and the EAS at 2 years (CA)

Domain score [a]	T2 at 24 mo (CA) mean (SD)	p-value	Difference mean (95% CI)	Adjusted p-value	Adjusted mean diff (SE) [d]	Cohen's d [b] (95% CI)
Cognitive		.17	4.5 (-11.1 to 2.0)	.02*	8.4 (3.5)	.66 (0.12; 1.20)
Intervention	101.7 (11.5)					
Control	97.2 (12.7)					
Motor		.04*	6.6 (-12.8 to -.49)	.01*	9.3 (3.4)	.85 (0.28; 1.42)
Intervention	103.6 (11.0)					
Control	96.9 (10.9)					
Gross motor subtest		.008*	1.1 (-2.0 to 1.1)	.01*	1.9 (.64)	.95 (0.37; 1.53)
Intervention	9.1 (1.7)					
Control	8.1 (2.0)					
Fine motor subtest		.10	1.1 (-2.7 to .39)	.20	1.1 (.83)	.55 (0.01 ; 1.11)
Intervention	12.0 (3.0)					
Control	11.0 (2.0)					

EAS (total scores) [c]	T2-24 months CA mean (SD)	p-value	Mean difference	Adjusted p-value	Adjusted mean diff (SE) [d]	Cohen's d [b] (95% CI)
Sensitivity		.16	1.5	.01*	3.0 (1.1)	.88 (0.24; 1.52)
Intervention	25.7 (3.3)					
Control	24.3 (3.4)					
Structuring		.38	.61	.02*	2.5 (1.0)	.71 (0.08; 1.34)
Intervention	25.9 (3.2)					
Control	25.1 (3.5)					
Nonintrusiveness		.72	.14	.09	2.7 (1.6)	.64 (0.01; 1.27)
Intervention	25.2 (4.4)					
Control	24.2 (4.2)					
Nonhostility		.35	.24	.48	0.5 (.60)	.29 (0.33; 0.91)
Intervention	27.7 (1.5)					
Control	27.6 (1.7)					
Child responsiveness		.66	.02	.05	2.2 (1.1)	.57 (0.05; 1.19)
Intervention	26.2 (3.3)					
Control	26.0 (3.8)					
Child involvement		.54	.54	.01*	2.8 (1.1)	.77 (0.14; 1.40)
Intervention	26.4 (3.3)					
Control	25.9 (3.6)					

[a] Scores on the BSIDIII-NL III are all corrected for prematurity.
[b] > 0.2 small effect, > 0.5 medium effect, > 0.8 large effect.
[c] EAS Scores ranging from 7 to 17 are considered to be Nonoptimally EA, 18 to 25 Inconsistently EA, and 26 to 29 Optimally EA.
[d] Adjusted for gestational age, Small for GA, gender and maternal education.

Discussion

In the Netherlands, a responsive parenting intervention (the TOP program) is implemented to support very preterm born children and their parents during 12 home visits throughout the first year after hospital discharge. The current pilot study was designed to explore the feasibility of an additional preventive intervention for very preterm born toddlers.

The additive intervention appears feasible because parents were willing to participate (response rate 64%), all participating parents completed the intervention, and the program was well received. In addition, the medium-to-large intervention effect sizes support our hypothesis that an additional preventive intervention improves responsive parenting and developmental and behavioral outcomes. We observed better quality of the emotional availability of the parent-child interactions in the intervention group. On the EAS, large effect sizes were found for the domain's sensitivity, structuring, and child involvement. This additional intervention seems to induce a better understanding of the toddler's behavior, potentially helping parents to match the information and support to the individual developmental opportunities and needs of their child. Sansavini et al. (30) described the relationship between parents and premature born children was mutual mother-infant co-regulation patterns and motor develop- with a shared, symmetric focus of attention. In line with Sansavini et al, we found a large intervention effect on motor development and a medium effect on cognitive development. Comparable early intervention programs do not report beneficial effects for cognitive development at 24 months of age (5, 14). In our previous studies on the effect of the IBAIP, positive effects on motor development were found at 24 months, whereas cognitive improvements were only found at 6 months and age 5 years (10, 12, 13) Our additive intervention may have boost the consistency of responsive interactions during the sensitive period of cognitive development. Our outcomes agree with the findings from Landry et al. (17) reporting that following intervention components that worked in infancy with a second dose of age-specific intervention strategies at toddler age achieves a better result.

In addition to the developmental improvements we found, parents in the intervention group reported fewer behavioral problems. Positive results on parental ratings of behavioral problems at 24 months (CA) also were reported in the Victorian Infant Brain Studies and Modified Mother-Infant Transaction Program study (5, 14) but not in combination with enhanced developmental outcomes.

Our outcomes may be more comparable with the studies of Treyvaud et al. (31, 32), who demonstrated that a more optimal home environment with engagement in shared activities and communication was associated with better cognitive development in preterm infants at age 2 years, including fewer externalizing and internalizing behaviors, and more regulated behavior and emotions. The developmental improvements as reported by parents on the ASQ-3 were only seen in the personal-social domain. The large improvements on the BSID-III-NL are not reflected in the parental perception of development in similar domains. In studies of the concurrent validity of the ASQ3 (33, 34), only a modest association between the ASQ-3 and the BSID-III-NL has been found. Furthermore, Steenis et al. (35) concluded that although the labels of the developmental domains are similar between the ASQ-3 and the BSID-III-NL, they do not seem to measure the same constructs. In addition, parental observation of developmental activities at home may differ from activities performed in a standardized test situation, causing differences between the scores of the ASQ-3 and the BSID-III-NL.

Only a small effect size for receptive language was found on the Lexi-list. This may point at the need for objective and more extensive instruments to measure communicative and language development to detect effects and to explore which intervention elements may be successful. Unfortunately, the BSID-III-NL, receptive communication subtest, was not available in Dutch at the time of the design of the study.

The results of this pilot study are promising, but outcomes should be interpreted with care. Our study group was small and not sufficiently powered to detect small differences between the groups. In addition, our study groups were not well balanced, requiring correction of our results by multivariate data analyses. Larger and more balanced study groups are necessary to confirm our moderate and large effect sizes. Thirty-four eligible families did not want to participate, and there was not enough background information on them to evaluate for possible inclusion bias. Nevertheless, we succeeded in including participants from a range of perinatal and sociodemographic backgrounds and various ethnic groups, consistent with the Amsterdam population.

Completing the questionnaires was difficult for some families, and therefore we were not able to collect all outcome measures in all families (Figure 4.1).

The BSID-III-NL and EAS were performed only at 24 months (CA) to minimize the additional appointments for the families. Therefore, we measured differences between the groups but not the changes over time. Future studies should include objective measures at baseline. We only measured outcomes

2 months after the intervention was completed and suggest that future trials should include a longer follow-up interval to evaluate for sustained intervention effects. Another limitation of this study was the use of questionnaires that are developed to detect developmental delay, rather than direct measurement of behavioral development. The questionnaires reflect parental perception of child development and child behavior, and parents were not blinded for treatment allocation, although investigators assessing outcomes were.

The positive results of this pilot study suggest that an additional preventive intervention at toddler age is feasible and potentially useful. The findings underscore recent insights that interactive dyadic processes with mutual adjustments support children's neurodevelopmental outcomes. This study adds that staged, age-appropriate interventions may boost the consistency of these responsive interactions and may further improve outcomes for preterm children. These promising results warrant further evaluation with a larger group to replicate the findings, confirm causality, assess intervention elements effecting change, and measure the long-term duration of intervention effects.

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Chapter 5

Parents' information needs during the first year at home with their very premature born child; a qualitative study

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Abstract

Objective: To obtain insights into parents' information needs during the first year at home with their very preterm (VP) born infant.

Methods: We conducted semi-structured interviews with parents of VP infants participating in a post-discharge responsive parenting intervention (TOP program). Online interviews were audiotaped and transcribed verbatim. Inductive thematic analysis was performed by two independent coders.

Results: Ten participants were interviewed and had various and changing information needs during the developmental trajectory of their infant. Three main themes emerged; (1) Help me understand and cope, (2) Be fully responsible for my baby, and (3) Teach me to do it myself. Available and used sources, such as the Internet, did not meet their information needs. Participants preferred their available and knowledgeable healthcare professionals for reassurance, tailored information, and practical guidance.

Conclusion: This study identified parents' information needs during the first year at home with their VP infant and uncovered underlying re-appearing needs to gain confidence in child-caring abilities and autonomy in decision-making about their infants' care.

Innovation: This study provides valuable information for healthcare professionals and eHealth developers to support parental self-efficacy during the first year after preterm birth.

Introduction

In the Netherlands, very preterm (VP) births before 32 weeks of gestational age (GA) account for 1.3% of live births and represent a proportion of 6.9% of all preterm births (< 37 weeks GA) (1). Unanticipated preterm birth and admission to the high-tech Neonatal Intensive Care Unit (NICU) are very stressful for parents and have a lasting impact (2-5). Parents have to cope with the emotional impact and consequences of preterm birth while trying to process information provided by medical staff about the infant's condition, treatment options, risks of complications, and uncertain long-term outcomes. Providing information is a key strategy since it generates a sense of control and facilitates the coping process (6). This in turn can improve self-efficacy, affection, and infant bonding, all of which are linked to positive parenting outcomes and long-term child developmental outcomes (7-9).

Different factors such as individual learning style, stress coping, the complexity of information, and communication skills of the health care professionals contribute to what extent patients can process and retain information (7). If VP parents have unmet information needs, they frequently use the internet to seek parentingrelated and health-related information. However, research has shown that the internet often lacks appropriate information for their situation (8, 12-18). Recent comprehensive literature searches revealed unaddressed information needs and support needs before and after discharge from the hospital (9-11). More promising results were found when using novel technologies such as smartphone applications designed around the needs of VP parents for the NICU and discharge period (19-22).

The Dutch healthcare system provides standardized and insurance covered follow-up programs for all VP infants. A responsive parenting intervention, the TOP program, is part of the standardized post-discharge care for infants born before 32 weeks of gestation and/or with a birth weight below 1500 g in the Netherlands and is provided by trained pediatric physical therapists (TOP interventionists) (23). The TOP program aims to enhance the developmental opportunities for VP infants at a critical time of their life by targeting parental responsiveness toward their child. A key strategy within the TOP program is to provide parents with information about their child's care and development and psychoeducation regarding the impact of premature birth (see Figure 5.1).

Most of this information is verbally provided by TOP interventionists during home visits and through an individualized written parental report. In our previous care evaluations, parents in the TOP program described difficulties in interpreting

their VP infants' behavioral responses, managing wake-sleep patterns, and consoling their infants. In addition, daily activities such as feeding, handling, and engaging were described as challenging. Consequently, parents sought confirmation about specific care decisions and other health and development related topics. We were interested in the possibility of using additional e-health technology to meet the information needs of VP parents. The aim of this study was therefore to gain insights into the VP parents' needs for information during the first year at home.

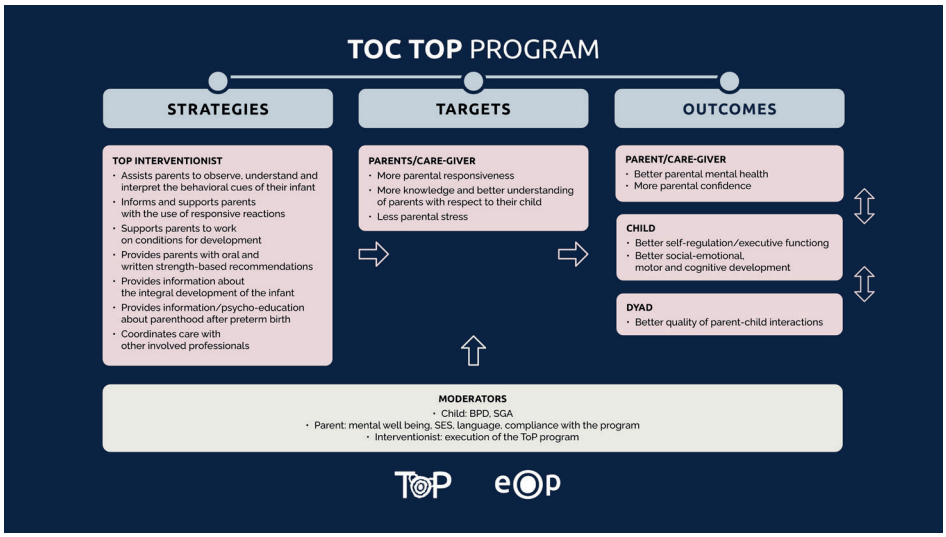

Figure 5.1. Theory of change of the TOP program.
Abbreviations: BPD, Bronchopulmonary dysplasia; SGA, Small for gestational age; SES, Socioeconomic status.

Methods

Design and setting

This qualitative study consisted of semi-structured individual interviews with VP parents. All participants were included in the Dutch follow-up program and received the TOP program and therefore had access to healthcare professionals such as a pediatrician and a TOP interventionist. Methods and results are reported according to the Consolidated Criteria for Reporting Qualitative Studies (COREQ) checklist (24).

Participants

This study was performed among Dutch parents. The inclusion criteria were parents who 1) received or just finished the TOP program; 2) had a VP infant with

the corrected age (CA) between 6 and 12 months; and 3) were Dutch-speaking. The criteria of CA > 6 months were set to cover sufficient time at home and also reduce recall bias. The research team informed the TOP interventionists about the aim and method of the study and asked them to approach families they were supporting with the TOP program. The purposive sampling was based on child characteristics: gestational age, first-born, multiple births, and parental factors such as gender, educational level, and family status (Table 5.1). The number of participants (n = 10) was expected to be sufficient to reach saturation and if necessary, could be expanded.

Data collection

An information letter was sent to parents who indicated interest in the study. Parents who consented to participate were contacted by telephone to schedule the interviews; the short online demographic questionnaire was subsequently sent by e-mail. The interviews were conducted by two interviewers (MF, RB) between April–June 2020, and lasted between 45 and 75 min. Due to the COVID-19 restrictions, participants were offered two options: by phone or virtual meeting. After verbal authorization, interviews were audiotaped. Interviewer MF, (female, nine years of experience working as a TOP interventionist) was involved as the main researcher in this study. Interviewer RB (female, pediatric physical therapist and clinical epidemiologist) had no pre-existing relationship with care for VP infants or their parents. The interview guide was developed in 4 phases 1) aligning interview questions with research questions; 2) constructing an inquiry-based conversation; 3) receiving feedback on the interview protocol; and 4) piloting the interview protocol as described by Castillo-Montoya (25).

The topics of the interview guide were based on a pragmatic literature search focusing on information for VP parents after discharge, promising e-health applications, and the use and preferences for information sources. Interviews were listened, to reflect on the interview style. The two interviewers had regular meetings to discuss the interview technique. Interview questions were open-ended regarding parents' personal experience. Only for topic 6 (requirements and preferences for online sources) the interviewers used more structured questions or provided examples. Asking about their current use of existing information apps, exploring what they liked or not liked helped participants to articulate their ideas and define their requirements.

The interview guide entailed six topics: 1) experiences with the transition to home and received or desired support; 2) needs for information regarding childcare and development during the first year at home; 3) information needs regarding parenthood; 4) perceptions of the provided information by TOP interventionists; 5) perceptions of using online resources; and 6) requirements and preferences for online resources.

Data analysis

An inductive thematic approach was based on the six-step process by Braun and Clarke (26). First (1), the transcribed interviews were checked individually (against the recorded interviews) and read by the first author to familiarize with the data. For the next step (2), data were coded and organized using Maxqda Analytics Pro 2020. Two researchers independently coded the first two interview transcripts and discussed differences in their coding. A preliminary codebook was made by EV and MF to code the remaining transcripts. The codebook was refined by MF as the analysis progressed and new codes emerged in the interviews. In the third phase (3), the initial codebook identified specific information needs as well as patterns, meaning, and essential elements underlying parental information needs. Codes were then collated into potential sub-themes. In phase 4, two researchers (MF and FN) reviewed, interpreted, and discussed the themes by analyzing the underlying latent meaning of the initial themes. Phase 5 identified the main three themes from the subthemes through ongoing reflective dialogue. The findings were then presented and discussed with the research team. In the report writing phase (6), the analysis and themes were related to the research question and current literature. Quotes were in this last phase translated by an official translator to contribute to the best possible representation (27).

Results

Participant characteristics

Seven mothers and three fathers were interviewed. All approached families expressed interest in participating and responded positively. Subsequently, they were scheduled for the interview. All ten participants received at least seven TOP intervention sessions (Mean (SD) = 10 (1.6) / range: 7–12), and all infants were > 7 months CA. The socio-economic background varied although parents without education or < 4 years after primary school were not represented (Table 5.1).

Table 5.1. Participants characteristics

	Relationship to infant	Age (year)	Marital status	Parental education*	Employment	First born	Infant GA Weeks/days	Singleton/ twin	Infant age (CA) **	# TOP home visits
1	Mother	28	Married	High	Paid job	First	26/0	Twin	12	12
2	Mother	27	Single	Middle	Paid job	Second	25/5	Singleton	8	10
3	Mother	25	Single	Middle	Sick leave	First	29/1	Twin	13	12
4	Father	41	Married	High	Paid job	First	24/6	Singleton	7	8
5	Mother	31	Married	High	Paid job	Second	31/3	Singleton	9	11
6	Mother	40	Married	High	Paid job	Second	26/0	Singleton	8	10
7	Father	30	Married	High	Paid job	First	31/1	Singleton	8	7
8	Mother	30	Married	Middle	Paid job	First	30/0	Singleton	8	9
9	Father	29	Married	Middle	Paid job	First	29/1	Twin	9	10
10	Mother	30	Married	High	Unemployed	First	25/1	Singleton	10	10

* Parental education; Low: no education/primary school/4 years after primary school; Middle: 5–8 years after primary school; High: bachelor/master level.
** Infant's corrected age during the interview.

The trajectory of information needs

On the first more general question, "How are you and (name infant] doing?", all parents immediately started with the conversation by looking back at the NICU experience and the period after discharge. When the interview questions guided them toward the preceding months, additional experiences and new information needs emerged. The main themes in their information needs were: 1) Help me understand and cope; 2) Be fully responsible for my baby; and 3) Teach me to do it myself. These themes re-appear during the first year at home (Table 5.2).

Table 5.2. Themes and subthemes

Themes	Subthemes		
Help me understand and cope	Making sense of the experience	Struggling to feel confident	Seeking information to comprehend
Be fully responsible for my baby	Ambivalent feelings	Loss of professional support	Seeking information for caregiving
Teach me to do it myself	Reassuring and regaining agency	Feeling competent and trusting own parenting skills	Needing personalized and specific information

Help me understand and cope

The theme 'help me understand and cope' appeared initially within the context of the NICU when participants sought information to regulate their distress and manage their uncertainties about their infants' well-being. As one parent described:

> "*Yes, and in one of those first weeks, eh…it became clear that my child had had a minor stroke, ehh… so, well you go and look that up too, while it applies to, in fact, 1 out of 3 or 1 out of 4 children have a stroke during the pregnancy but you don't know that.*" [P9]

Although the information from NICU professionals was generally perceived as adequate and prompt, all our participants used the internet as an additional information source to familiarize themselves with the NICU environment and their infant's medical condition. The search for information on the internet was described as: difficult to find, not helpful, and too many horror stories. These first negative experiences with internet search influenced further use of

online sources and often led to the avoidance of the internet during the first year at home.

> "*Yes, well every child is different. You can Google things, but I have to say it didn't help me much at all, it was extremely difficult for us to find information on the Internet, so, in fact, at one point we simply stopped trying.*" [P10]

Participants expressed increased feelings of insecurity in anticipation of being discharged home. They all expressed mixed feelings of joy and readiness to go home while simultaneously having the awareness and fear of being fully responsible for their vulnerable child.

> "*You're pleased to be home again, but we also found it extremely tiring to be back home.*" [P1]

Once at home, parents encountered new challenges such as poor feeding intake, sleeping problems, and regulating their infant and expressed their insecurity and need for information for baby care.

> "*How could I know if there are too many stimuli? So, when do I give a bath, what can my baby handle, that is what I needed to know.*" [P1]

After the first few months, parents became more aware of the ongoing risks of prematurity and neurodevelopmental delays and sought information to compare their VP-born children with existing norms for full-term infants.

> "*With a premature baby, of course, you use their corrected age, uh… so you can do that for the first two/three months because you know that they're nine weeks behind schedule, but will they catch up?*" [P10]

Other participants started processing the impact of preterm birth on their well-being and sought support from professionals (psychologists) or found support from the parents' association Facebook group.

> "*How do you keep yourself upright and not get buried under the weight of taking care of your baby? This can be hard on your relationship as well, how do you share the responsibilities?*" [P1]

> "*The Facebook group is made up of parents with premature babies or babies who were ill at birth. It's a nice group.*" [P4]

Be fully responsible for my baby

Participants perceived the transition to becoming a parent in an institutional context as complex. Feelings of insecurity and challenges associated with their parental role continue to be present in the transition phase of going home and throughout the first year. All of our interviewed parents described the fragile sense of parental agency and insecurity about being fully responsible for their vulnerable child.

> "*Yes, because after all, you're suddenly responsible for everything, whereas in the hospital you always had a safety net. You don't have that at home. Suddenly, find yourself without that safety net, in a different environment where you suddenly have to do everything yourself; well that is simply a whole new ball game.*" [P6]

Participants expressed conflicting feelings about holding on to prescribed schedules and protocols and starting to trust their parenting skills.

> "*The same goes for growth, they have to grow. We had weighing scales at home for a time. But after a while, we got rid of that too. We decided we had to let it go and keep a close eye on the baby, decide if it was feeling comfortable.*" [P3]

Engagement in their infant's hospital care improved their practical skills such as diaper change and provided them with insights about their infant's health issues. Some parents used the possibility of a 24-h presence (rooming-in), to prepare for discharge. For other parents, the transition to home came unexpectedly fast. Despite the preparation, parents experienced a knowledge gap between hospital care and care at home.

> "*I found that really quite overwhelming. And then you're back home, and of course, before you go home the nurse does give you information, but it's more the basics, like how to get a bottle ready and test the temperature and so on, but very little else. Of course, in the end, you have to work it out for yourself. You have to get to know your child. But in general, I received very little support during that period.*" [P4]

In the hospital, information on the growth and body functions of the infant is collected daily by highly sophisticated equipment or health care professionals. The absence of a healthcare team to rely on, feeling responsible, and making caregiving decisions was challenging. Parents expressed difficulties in transfer-

ring their acquired skills and knowledge into their new situation and described new emerging information needs about practical topics, such as diapering, bed making, feeding, recognizing their preterm infant's needs, and sleep and wake patterns for the first days at home.

Teach me to do it myself

To achieve the desired confidence, parents require experienced, knowledgeable professionals who interact sensitively with the parent's needs and can reassure them.

> "*Like with postnatal maternity care, for example. The midwife was really good, it was just that she didn't know much about premature babies. I think it must be great if you have someone with experience in this field.*" [P1]

Parents appreciated the accurate timing and dosage of information provided by the TOP interventionist. This practical and deployable information was described as transferable and seemed to enhance their independence.

> "*If I needed advice about sleeping, for example, I was given information about sleeping. I didn't get too much information that wasn't relevant. The program was very much tailor-made. And that's what you want, that it suits your baby's needs. That you can use it in your situation.*" [P1]

During the course of the first year, participants' information needs changed to a need for specific and personalized information. Depending on the infants' developmental trajectory and own path of processing, participants identified their concerns and formulated their ideas for an additional information source. The ten most relevant topics, which parents sought more information about, are outlined in Table 5.3.

There was a high agreement about these topics between parents. Parents indicated either had searched for this information on the internet or had missed it. Participants desired knowledge on how they can contribute to their infants' development, this was sometimes reinforced by concerns about the long-term impact of the premature birth.

> "*But I'm starting to get just a bit worried about things like, hmm, what about if my child is not doing so well when it starts school? I want her to have a problem-free life as possible. What can we do to make sure that happens?*" [P7]

During the interviews, parents reflected on which additional information they would have liked to have and in which period or situation. All parents first and foremost seem to benefit from a comprehensive and tailored source accessible at desired times.

> "*You don't need all the information upfront, but you need to know more about where you can run into and who can I then ask for advice or where can find the information.*" [P1]

Table 5.3. Topics and subtopics

TOPIC	Subtopics and concerns identified by participants
General health issues	Common pediatric health issues such as fever, a cold, getting teeth. How to decide if I need to visit the pediatrician or a general practitioner? Do I need to handle it differently with a more vulnerable baby?
Motor development	Information about motor milestones (small steps), sequences in motor development. And how is this different for premature infants.
Regulation	Sensory input; what can my premature baby handle? Co-regulation: what can I do to help my baby to regulate?
Feeding	Basic information about formula, letting go of tight schedules, and transitioning to solid foods—digestion-related questions.
Sleeping	How do I recognize the patterns of sleep and my baby's fatigue? Suggestions for how to put your baby to sleep. Is it bad to let my premature baby cry?
Prematurity	Why and until when use the corrected age? What are the anticipated problems for premature infants?
Long term outcomes	What are the increased risks for long term problems and what to expect for my baby? What can we do right now to prevent adverse outcomes?
Parenthood	Impact of premature birth and intensive caregiving at home on relationships and own wellbeing. How do you keep balance?
Going back to work	What if my baby is not ready for daycare? How do I inform my manager?
Professionals	What kind of help is available? Where do I find a knowledgeable professional?

Discussion and conclusion

Discussion

This study was set up to obtain insight into VP parents' needs for information during the first year at home. Family-centered care (FCC) and the European standards for newborn health are extensively implemented in the Netherlands (28). The participants in this study appreciated the efforts to involve them in hospital care and prepare them for discharge. However, this study clearly showed that despite these efforts, parents need personalized and specific support and information for new emerging developmental phases of their infant. Participants expressed the need for practical and confirming information to be confident in taking care of their baby at home. Knowledgeable and specialized healthcare professionals, such as the TOP interventionist and the pediatrician, were considered the most important source of information. Parents stated that reliable and tailored online sources can complement post-discharge care, but should not replace the professional. This is in line with a scoping review from David-Strauss and colleagues who highlighted the need for in-person support, such as a specialized home visitor, in addition to online sources (9).

The NICU period appeared to have a notable negative lasting impact on gaining self-confidence, making autonomous decisions, and trusting their parenting skills. Our participants seemed to benefit from the practical guidance to learn about their infants' behavior and needs, positive appraisal, and confirmation of their capabilities. Our findings correspond with Bandura's self-efficacy theory (1977) which describes the development of a strong sense of efficacy as influenced by four resources: mastery experiences, vicarious experiences, social persuasion, and somatic and emotional states (3, 29).

The three ultimate themes, namely 1) Help me understand and cope; 2) Be fully responsible for my baby; and 3) Teach me to do it myself, are strongly interconnected to each other and re-appear during the first year after preterm birth at three distinctive periods; hospital, discharge, at home. These identified underlying needs are in line with the parental experiences after discharge described in the meta-synthesis by Adama et al. (31) and the phenomenological study by Murdoch and Franck (32). This qualitative study adds new insights into underlying requirements for reassurance, trajectory-dependent topics, and changing practical information needs. These insights could improve not only existing discharge programs but should also be considered as foundations for creating new intervention programs.

Overall, participants perceived the available information about the impact and consequences of VP birth as insufficient or not appropriate for their situation and indicated ten important topics for the first year at home corresponding with the priority topics found by Alderdice (33). Parents were less likely to use online sources after hospital discharge, shaped by their negative experience of retrieving helpful information. This lack of written (discharge) information to complement interpersonal exchanges was also described by Lorie in a qualitative study with Dutch (NICU) parents (34). This underlines the need for the joint development of an online information source for VP parents, growing with them during the different phases of their trajectory.

Developing a tailored information source for VP parents at home is a first and essential step before evaluating if e-health technology can be a good strategy to support parental self-efficacy. In this study, we asked participants to reflect on this future scenario and learned it is not easily solved by providing just more information to enhance knowledge. To make the use of (online) information sources successful for VP parents, we must also take into account the complexity of the found underlying emotional needs, all in line with how Lee described self-efficacy; a person's belief in his or her ability to succeed in a particular situation (30). Also endorsed by van Gemert-Pijnen who described the need for a comprehensive and holistic view of supporting health care by e-Health technology addressing the need for recognizing interdependencies between individual characteristics and the ability to use e-Health technology (35, 36). When carefully considering the unique needs of the VP parents, the combination of e-health technology and a healthcare professional can support the shift from time-dependent and healthcare professional-dependent delivery of information to self-management.

Limitations

The number of participants (10) was set in advance and followed by strategic sampling. The interview number was large enough to reach data saturation. All participants had a good understanding of the Dutch language, which was a pre-condition for the interviews but may have limited the inclusivity of parents with a low educational level. Involving non-native Dutch parents and parents with a lower educational background could have provided an additional layer of understanding. However, the middle and highly educated participants expressed their information needs, and we expect that the parents with lower education will also require more tailored information. Further research in the development

of information sources is essential to not enlarge the already existing disparities between education levels.

Innovation

This study shows the importance of retrieving in-depth information about parents' needs before designing a useful and parent-empowering e-health application. Our findings target unaddressed needs in existing (digital) information sources for VP parents after discharge. By addressing the underlying themes found in this study, e-health technology solutions can be more tailored for the unique needs of the target group. This study is useful because it gives information that can be directly converted into practical implementation. Participants did not only reflect on their trajectory dependent information needs but also provided ideas for improvement. The underlying need to feel confident and gain autonomy requires strength-based and re-assuring information and tone of voice. Participants identified information needs for a substantial number of topics e.g. infant feeding, sleeping, or motor development. The information needs change over time, requiring specific age- and development-related information provision along the trajectory of the first year. The transferability to their own unique situation requires information that is easy to understand and apply. Another important finding was the preferred approach for support from a professional in combination with a digital information source (blended care). Findings of this study can be used as a starting point for development and co-designing an applicable and useful information source to support VP parents.

Conclusion

The present study shows the pervasive impact of premature birth on parental confidence and identifies parental information needs during the first year at home with their VP infant. The underlying themes further extends the understanding of how to support parental needs for information. Participants expressed a preference for a knowledgeable healthcare professional in combination with a customized information source accessible when needed. These results are relevant for e-health developers in the field of neonatal care since these findings give direction for the (content) development and use of e-health technology to support VP parents' self-efficacy.

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Chapter 6

Evaluating mobile information applications for parents of preterm infants after hospital discharge: A systematic app review

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Abstract

Background: After hospital discharge, parents of preterm infants need accessible and reliable information to gain confidence and skills in their child-caring abilities and parental autonomy. Parental need for information after hospital discharge includes topics related to prematurity, such as crying, feeding, sleeping, infant care, general health, and neuromotor development. However, parents report difficulty in finding and understanding this information. Mobile apps have the potential to improve information provision.

Objective: The aim of this systematic app review was (1) to identify mobile apps for parents of preterm infants targeting the period after hospital discharge and (2) evaluate the content, quality of the app, and understandability and actionability of the information material.

Methods: We systematically searched for apps in the Apple App Store, Google Play Store, and Google along with a literature search using PubMed. Multiple keywords were used (e.g. “preterm baby”, “app”, and “home”). Apps were included when they provided information for parents on topics and content related to prematurity after hospital discharge. To examine app content related to the post-discharge period, apps were reviewed and topics were identified. The Mobile App Rating Scale (MARS) was used to measure the app’s quality, and the Patient Education Materials Assessment Tool for Audiovisual Materials (PEMAT-AV) was used to measure the understandability and actionability of the information material.

Results: After the initial search, the title and descriptions of 202 apps were screened for eligibility. Eventually, nine English apps were included in the review. Information related to the post-discharge period constituted only a small part of the app’s content. Most commonly addressed topics related to the period at home were vaccinations, follow-up, feeding, and using home oxygen. Using the MARS, only one of nine apps received a good score for overall quality (“My preemie app”) and seven apps received an acceptable score. Only four apps scored high on Understandability of the PEMAT-AV, and six apps scored high on Actionability. No Dutch apps were identified.

Conclusions: The current availability of mobile information apps for parents of preterm infants targeting the period after hospital discharge is limited. Nine English apps were identified, that contained a small portion related to the post-discharge period. This content is not comprehensive for the post-discharge period: topics indicated as relevant by parents such as crying in preterm infants, diaper change in preterms, or parental well-being after preterm birth are often missing. The overall quality of the apps is only acceptable. Although the reliability of the information was close to good, the understandability of the apps was moderate. Recommendations for future app development include more relevant and understandable information related to the post-discharge period, that meets the demand of VP parents.

Introduction

Yearly, 13.4 million babies are born preterm (PT) (< 37 weeks of gestation), of which 2 million infants are born very preterm (VPT) (< 32 weeks of gestation) (1). VPT infants are frequently discharged from hospital prior to their term date (2), and parents often feel inadequately prepared to take their vulnerable infant home (3). Without the continuous professional support of the Neonatal Intensive Care Unit (NICU), parents lack confidence or competence in infant caregiving. Parents report practical, emotional, and financial challenges at home and the need for practical support regarding baby caregiving tasks, feeding, medication, or managing unexpected health issues (4). The uncertainty about health, growth, and neuromotor development of their VPT infant can heighten parental anxiety and negatively affect parents' caregiving behavior (5).

Parental confidence and competence in caring for their VPT infant can be increased by professional support and tailored information and is thus an important approach to improve parental and infant outcomes (6, 7). Caring for a VPT infant after hospital discharge can be more demanding for parents than caring for a typically developing infant. VPT infants show different behaviors such as reduced activity, alertness, and responsiveness that require specific parenting skills to interpret their baby's cues (4). Parents therefore require information on common topics specifically targeting prematurity, such as crying (how to comfort a preterm infant), feeding (how and when to transition to solid foods), sleeping (recognize pattern of sleep and fatigue in their baby), infant regulation (help their baby to regulate), infant care (diaper change in a very small infant), general health (when to contact a pediatrician), and neuromotor development (differences in milestones between term and preterm infants) (5, 8). This information specific for premature infants is, however, not available on the internet (6). Parents appreciate that general parenting websites provide accessible information on newborn topics such as feedings and digestion, but the content is perceived as less appropriate for parents with a VPT infant (6). Therefore, practical and tailored information is necessary to increase knowledge and skills to support parents to feel confident in taking care of their preterm baby at home. To accommodate their underlying emotional needs, parents prefer information that is strength-based and confirming or reassuring in their caregiving (8).

Almost all parents in the NICU use their smartphone to search for information regarding prematurity on the internet (9). For instance, in the Netherlands

information is provided by the Dutch parent organization (Care4neo) (10). Facilitated by the ease of use, the 24/7 availability and ability to make information attractive, mobile health (mHealth) apps are promising tools to provide health information (11).

In general, parental knowledge about infant development is associated with better caregiving behavior and improved infant development (12). Mobile apps have the potential to improve parental well-being and parenting in the perinatal period (13). Mobile health apps vary in quality, but many are of moderate quality or out of date (11, 14). A previous review on information apps targeting parents with an infant that was still admitted to the NICU showed that only a quarter of the apps for parents were considered of good quality (15). For optimal support, parental needs for information should be incorporated in the content of the app (16). Parents have ongoing information needs, but what they want to know changes over time (8). After discharge home, when hospital staff support is lacking, parents need different information to feel competent in their caregiving than during their initial hospital stay.

For health care professionals and parents, it is important to be able to use high quality apps, including the engagement, functionality, aesthetics, and information quality. Therefore, information content of mHealth applications needs to be understandable for all parents, irrespective of health literacy levels. Since preterm birth has been consistently associated with lower socioeconomic status (17), also low health literacy is a prevalent issue in parents of VPT infants. Health literacy refers to the skills needed to function effectively in the health care environment (18) and low health literacy is associated with poorer use of health care services and poorer health outcomes (18). Parents with lower health literacy may encounter difficulties in obtaining, processing, using, and interpreting information in mHealth applications (19). To benefit from mHealth, parents require digital health literacy skills, such as using digital devices, searching for and understanding information, and evaluating the validity of the information (20). However, to date, little is known about the quality, understandability, and actionability of available mHealth applications designed to support parents of VPT infants after hospital discharge. Therefore, the aim of this app review was 1) to identify mobile information apps for parents of preterm infants targeting the period after hospital discharge and 2) to evaluate the content, quality of the app, and understandability and actionability of the information material.

Methods

Study design

This systematic review of mobile apps followed systematic review methodology adhering to Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) standards (21) and published conduct and reporting recommendations for systematic app store reviews (22).

Search strategy

To ensure the identification of relevant mobile apps, a comprehensive search was employed, using four different strategies. Apps were directly searched in 1) Apple App Store for iOS, and 2) Google Play Store for Android. In addition, mobile apps were also searched via 3) Google and 4) PubMed. The search in the app stores and Google was performed in December 2023. The search in PubMed was performed in May 2022. In the first three search strategies, keywords were used both in Dutch and in English.

The different search machines implied different search strategies. In the app stores, separatkey terms (in Dutch and English), were used in the search field: “preterm baby”, “preemie”, “premature”, “NICU”, “discharge”. For the Google internet search, the term “app” was always used and combined with terms “preterm baby”, “preemie”, “prematurity”, NICU”, “Neonatology”, “incubator”. The PubMed search combined terms “parent”, “mother”, “father” or “caregiver” AND “premature birth”, “premature infant”, “preterm”, “prematurity” AND “mobile application*”, “smartphone application*”, “health app*”, “mobile app*”. No MESH terms were used for the PubMed search. The aim of the Google internet- and PubMed search, was to identify more apps that were subsequently retrieved from one of the App Stores.

The search in the Apple App Store and PubMed provided a certain amount of apps and papers. These were all reviewed for eligibility. In the Google Play Store, the search yielded a continuous stream of apps, many of which were not relevant to our inclusion criteria. Therefore, we limited the screening to the first 50 apps that were displayed in the search results. These are typically ordered by relevance and popularity and align with how parents would conduct such a search. In the Google web search, the first 2 pages of results were reviewed to evaluate whether an app for parents of preterm infants was described. Search was ended when 2 pages did not contain new hits. Limiting search results is a common practice in app and website reviews, as later results are less likely to

be accessed by parents, often align less with the search criteria, and parents are unlikely to continue their search beyond a certain point (11, 15, 23).

App selection

Several in- and exclusion criteria were used to select the mobile apps (Table 6.1). The free of charge criterion was used because we wanted to ensure that apps were available to all parents regardless of their socio-economic status, income or willingness to pay for a mobile app. After removing duplicates between the two app stores, the Google and PubMed search, the app descriptions and features were first screened in the Apple App Store or Google Play Store by one researcher (RG) and discussed within the research team for eligibility. Apps that fulfilled the inclusion criteria were then downloaded. Two reviewers (RG and MJ) screened the apps for inclusion in the full app review and discussed the eligibility within the research team.

Table 6.1. Inclusion/exclusion criteria for app selection

Inclusion criteria	Exclusion criteria
Topic	
VPT infants	Typical developing infants
Timing of information	
Period after hospital discharge	Only during hospital stay
Language	
English or Dutch	Other languages than English or Dutch
Access	
No access code	Access code required
Information in the app	
Directed to parents	Not directed to parents
Download	
App available for download	App not available for download
Charge	
App free of charge	Paid app

Data extraction and quality assessment

For each app, the following data was collected: name of the app, operating system, developer and its affiliation, language, target population, year of last update, and a brief description of the app. To evaluate the post-discharge

hospital content, a list of topics per app was created. To evaluate the quality of the apps and the understandability and actionability of the information material, two independent reviewers (MJ, RG) trained themselves to use the Mobile Application Rating Scale (MARS) (24) and the Patient Education Materials Assessment Tool for Audiovisual Materials (PEMAT-A/V) (25). Thereafter, all included apps were independently evaluated by the two reviewers and disagreements were resolved until consensus was reached. When no consensus was reached, the research team was involved. For each instrument, a structured data retrieval form was composed, using a spread sheet in Microsoft Excel.

Mobile Application Rating Scale (MARS)

The MARS is a tool for assessing the quality of mobile health apps. The MARS consists of four objective scales: "engagement" (5 items: fun, interesting, customizable, interactive, well-targeted to audience), "functionality" (4 items: app functioning, easy to learn, navigation, and gestural design), "aesthetics" (3 items: layout, graphics, and visual appeal), and "information quality" (7 items: accuracy of app description, measurable and achievable goals, quality of information, quantity of information, visual information, credibility, evidence based). Each item is rated on a 5-point rating scale, ranging from 1 "inadequate" to 5 "excellent". Each item has specific descriptions for these rating anchors. Some items have the option "not applicable". In addition, there is one scale for "subjective quality" (4 items: recommendation of the app, estimated frequency of use, willingness to pay, overall star rating of the app). The first three items are rated on a 5-point scale, and the last item on a 3-point scale. The overall mean score for the four objective subscales is calculated, excluding the items rated as not applicable. The MARS has a high internal consistency (alpha = 0.90) and high interrater reliability (ICC = 0.79) (24). For this study, we used the Dutch version of the MARS (25).

The Patient Education Materials Assessment Tool for Audiovisual Materials (PEMAT-A/V)

The PEMAT-A/V is an instrument that assesses the understandability and actionability of audiovisual patient education materials (26). The PEMAT-A/V consists of two scales: understandability (13 items) and actionability (4 items). Understandability is defined as the ability of people from diverse backgrounds with varying levels of health literacy to comprehend educational material and extract key messages. Actionability is defined as the ability of learners to identify what

actions can be taken on the basis of educational material information. Understandability includes 19 items evaluating the content, word choice and style, number usage, organization, layout and design, and use of visual aids. Actionability contains four items and evaluates whether the material 1) identifies an action the user can take; 2) the user is directly addressed; 3) breaks down an action into manageable steps; and 4) explains how to use the charts, graphs, tables or diagrams to take action. Items are rated with "disagree" (= 0 points) or "agree" (= 1 point). Some items have the additional option "not applicable". The PEMAT is designed to be completed by professionals and helps them to select education material that is understandable and actionable. The PEMAT items are based on other instruments and concepts for developing education material and reliable for raters not trained in the use of the PEMAT. The researchers read the information in the apps and considered each item from a parental perspective, specifically a parent with low health literacy skills. The researchers did have experience with developing information for people with low health literacy skills. The scores for the two scales are calculated as a percentage, ranging from 0–100. A higher score reflects more understandability or actionability. An expert panel established the face and content validity. Interrater reliability was moderate according to Cohen's kappa (0.50), but with a high absolute agreement of 80% and high agreement when calculated by Gwet's AC1 (0.71). Internal consistency was strong, (Cronbach's alpha = 0.76) and the average item-total correlation = 0.62. Construct validation was established based on differences in actionable and poorly actionable material as well as a strong negative correlation between grade level and both consumer-testing results and PEMAT scores (26).

Data analysis

Data analysis was performed using IBM Statistical Package for Social Sciences software (IBM SPSS), version 26. The MARS item scores were averaged for the engagement, functionality, aesthetics, and information subscales. These scores for app quality were then averaged, creating a mean (SD) app quality score. Descriptive statistics were used to summarize the results of the MARS and the PEMAT-A/V. To evaluate consistency between raters, the intraclass correlation coefficient (ICC) between the raters was calculated for the MARS and the PEMAT-A/V. Rater agreement was examined by ICC based on a two-way mixed-effects model. An ICC of < 0.50 is considered poor, 0.51–0.75 as moderate, 0.76–0.89 as good, and > 0.90 as excellent.

Results

Search results

The search yielded 191 apps in the Apple and Google Play stores, and additionally 36 apps in Google and PubMed. After removing duplicates, 185 apps remained (Figure 6.1). Based on title and description in the app stores, 169 apps were excluded. The majority of the excluded apps did not contain information on preterm born infants (N = 99; 58%), did not target parents but health care professionals (N = 29; 17%), or did not contain information but for instance only growth diaries (N = 6; 4%). Only one app was excluded because it was a paid app. Twelve apps (6%) that were identified via Google or PubMed could not be retrieved anymore in the app stores. The remaining 16 apps were downloaded and screened for inclusion in the full app review. One app was not available for downloading. Finally, 9 apps fulfilled the inclusion criteria and were included in the final analysis. The majority of the apps was available in both app stores (N = 5), three apps were only available in Apple App Store and one app was only available in the Google Play Store. Two apps were also described in a scientific paper. One paper describes the content of the MyPreemie app, based on an earlier book Preemies: the Essential Guide for Parents of Premature Babies, supplemented with new tools (27). The co-design approach of the Preterm Connect app has been described across three settings with different socially, economically and culturally participants (28). The preliminary findings show similar parental needs, but different preferences across the study populations.

Characteristics of the apps

The apps were developed in the United States of America (n = 4: A2, A4, A7, A9), the United Kingdom (n = 2: A3, A5), Australia (n = 1: A6) and New Zealand (n = 1: A1), and South Africa (n = 1: A8) (Table 6.2). All nine apps were in English, no Dutch apps fulfilled the inclusion criteria. One app (A2) had information in 25 languages and another app (A4) was also available in Spanish. The apps were developed by reputable sources, including hospitals (n = 4: A1, A3, A6, A7), non-governmental organizations (NGO) (n = 3: A2, A4, A5), and universities (n = 2: A8, A9). The last update of the app varied between 3 weeks and 3 years, with the majority of the apps updated more than a year ago. The size of the apps varied between 7.2 and 102.2 MB. Three apps were downloaded more than 1000 times (A1, A2, A4) and received positive ratings ranging from 4.0 to 4.9 on a scale from 0–5.

Table 6.2. General characteristics of the apps for parents of preterm infants after hospital discharge

ID	Name	Op. system	Country	Version	Last update	Target population	Developer	Affiliation	Brief description	Language	Size (MB)	Rating	Downloads
A1	Babble NZ Neonatal Family App	Apple IOS, Android	New Zealand	2.7.1	2021	Parent with a baby in the NICU	Neonatal unit at Midcentral Health	Hospital	A reliable source of information about the NICU.	English	7.2	4.9 (N = 10)[a] n.a.[b]	1k+
A2	Birth & Beyond	Apple IOS, Android	USA	1.16	2022	Mothers of newborn babies	Global Health Media Project	NGO	48 videos in 30 languages.	25 languages	10.7	4.8 (N = 26)[a] n.a.[b]	10k+
A3	Family Delivered Neonatal Care (IFDC)	Apple IOS	UK	1.1.11	2023	Parents in the NICU	Imperial College healthcare NHS trust	Hospital	The app offers up to date and comprehensive educational material, developmental timeline and diary functions to document the neonatal journey.	English	29.2	n.a.[b]	n.a.
A4	MyPreemie app	Apple IOS, Android	USA	2.3.1	2020	Families of premature babies	Graham's foundation	NGO	Toolkit for the practical and emotional needs of families of premature babies.	English/ Spanish	35.4	4.0 (N = 182)[a] 5 (N = 1)[b]	10k+
A5	My Prem Baby	Apple IOS	UK	1.8.1	2023	Parents of a premature baby	Tommy's	NGO	Track and monitor the journey with your premature baby.	English	50.9	n.a.[b]	n.a.

ID	Name	Op. system	Country	Version	Last update	Target population	Developer	Affiliation	Brief description	Language	Size (MB)	Rating	Downloads
A6	Neonatal Care and Me	Apple IOS, Android	Australia	n.a.	2021	Parents of a baby in the NICU or special care nursery	South Western Sydney Local Health District	Hospital	Tool while the baby is in the hospital and beyond and whilst being guided by health professional.	English	102.2	n.a.[a] n.a.[b]	n.a.
A7	Our Journey in the NICU	Apple IOS	USA	2.1	2020	Families of children in the NICU	Phoenix Children's hospital	Hospital	Identify what families need to know before taking their child home from the hospital.	English	8.2	n.a.[b]	n.a.
A8	Preemie Mom Care	Android	South Africa	2.0.5	2020	Mothers of hospitalized premature infants	UCT Human Computer Interaction lab	University	Provides supportive information to mothers of hospitalized premature infants as they partake in the care of their infant.	English Afrikaans Xhosa	24.8	n.a.[a]	500+
A9	Preterm Connect	Apple IOS, Android	USA	1.8.5	2023	Parents of a preterm baby	Chih H Wang	University	Connect with other women through forms for preterm birth. Articles and videos about caring for a preterm baby and yourself.	English	63.5	n.a.[a] n.a.[b]	50+

[a] Rating in Google Play; [b] Rating in Apple App Store (range 0–5); n.a., not available.

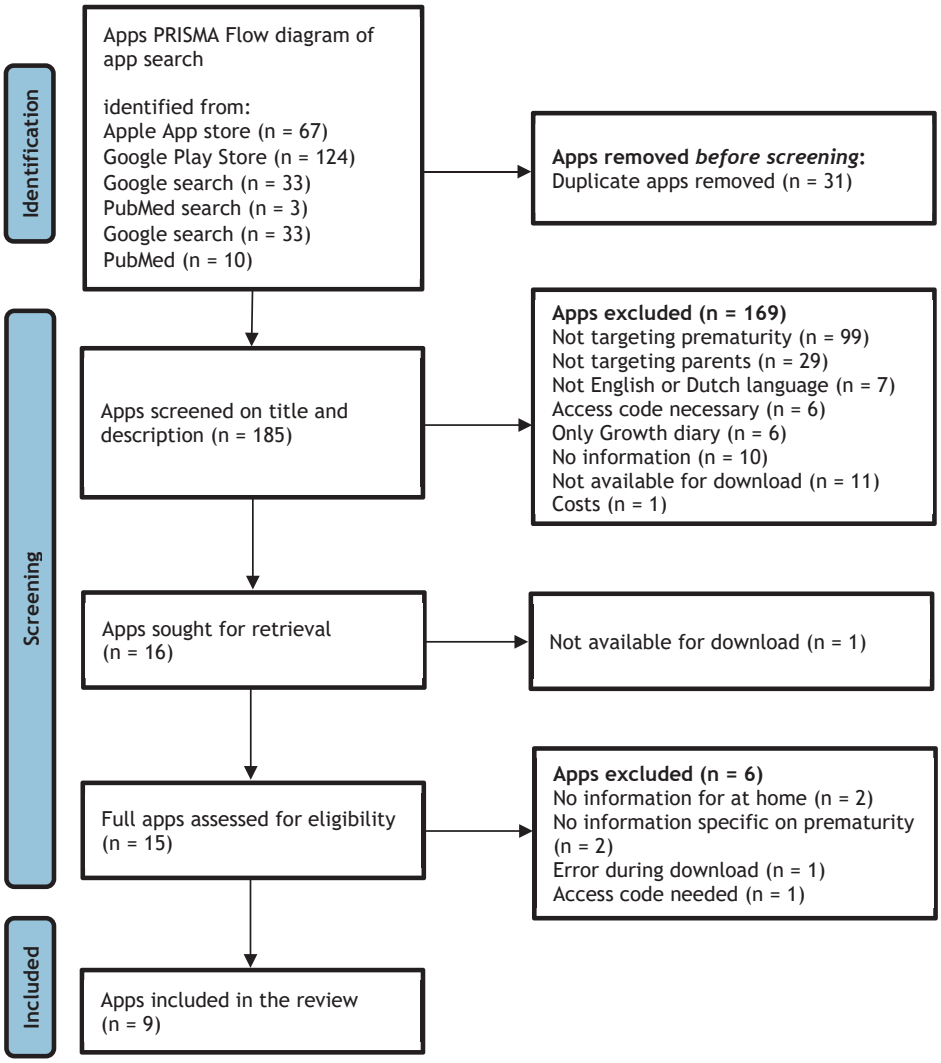

Figure 6.1. PRISMA flow diagram of search for apps for parents of preterm infants after hospital discharge.

App content

Most of the information in all apps was directed to the period in the NICU. The quantity of information for the post-hospital discharge period was limited. Some apps have one "chapter" that covers the post-discharge period (A5: "at home with baby"; A9: "Parenting at home") whereas other apps have subthemes within a chapter (A4: "Preemie Parenting", "going home"). Topics that were addressed varied widely between the apps (Table 6.3). The most common topics

that were covered in the apps related to the period post-discharge was: vaccinations, follow-up, and recognizing signs of illness. More practical information was provided on feeding, using home oxygen, and sleep (Table 6.3). Less often, the apps provided information on aspects that were reported as relevant by parents (5, 8) as diaper change (A5, A9), crying (A1, A7, A9), or parental well-being (A4, A5, A9).

Besides information provision, the apps included also other functionalities, including monitoring and tracking of infants' weight and height, amount and duration of feeding, or parental mood (A4, A5, A6, A7) (Table 6.3). The option of making notes was also provided by four apps (A3, A4, A7, A9). Sharing information from the app with others was available in three apps (A2, A4, A5). Community groups were only incorporated in one app (A9).

Table 6.3. Most common post-discharge topics and functionalities of the apps

Common topics post-discharge	Number of apps	App IDs
Feeding	7	A2, A3, A4, A5, A6, A8, A9
Vaccinations	6	A1, A2, A3, A4, A5, A9
Follow-up	6	A1, A3, A4, A5, A8, A9
Signs of illness in baby	5	A1, A2, A7, A8, A9
Sleep	5	A1, A3, A5, A7, A9
Home oxygen	4	A3, A4, A5, A7
Bathing	4	A5, A6, A7, A9
Functionalities		
Monitoring or tracking (diary, growth, weight)	3	A4, A5, A6
Making notes/saving questions	3	A4, A5, A7
Sharing information	3	A2, A4, A5
Saving articles	3	A1, A2, A9
Community groups	1	A9

Quality of the apps (MARS)

The interrater reliability of the MARS of the two raters was high (ICC = 0.99, CI 0.98–0.99). The overall mean quality (range 0–5) of the 9 apps was 3.4 (SD 0.5; range 2.3 to 4.3) (Table 6.3). The majority of the apps (N = 7) scored between acceptable to good, one app (A7) scored below acceptable, and only one app (A4) scored above good. There was a difference in the ratings between the four objective MARS scales. Engagement was rated as poor to acceptable (2.8 (0.6), range 1.8–3.4), specifically due to low scores on entertainment, customization, and interactivity. The aesthetics domain was acceptable (3.2 (1.0), range 1.7–5).

Information quality and functionality were close to good, (3.8 (0.6) range: 2.3–4.3), and (3.9 (0.5), range 3.3–4.8) respectively. Several apps received a good score (> 4) for information quality (A2, A4, A5, A6, A9), functionality (A2, A3, A4, A8), aesthetics (A4, A6), and overall mean quality (A4). None of the apps received a good score for engagement. The subjective quality (total range 0 to 18) ranged from 8 to 16, with a mean of 12.6 (3.0). Seven apps (A1, A2, A3, A4, A5, A6, A9) received a good score for subjective quality.

Understandability and actionability of the apps (PEMAT-AV)

The interrater reliability of the PEMAT-AV of the two raters was high (Understandability: ICC = 0.89, CI 0.55–0.98; Actionability: ICC = 0.91, CI 0.59–0.98). The mean (SD) understandability of the apps was 78% (12%); ranging from 55% to 100% (Table 6.4). Only one app (A2) scored the maximum of 100% for understandability. Lower ratings were obtained when lacking a summary of the information or visual cues to draw attention to key points. The mean actionability was 85% (24%; range 33% to 100%). Lower ratings were obtained when not addressing the user directly, or not breaking the action down into manageable, explicit steps. Six apps (A2, A3, A5, A6, A7, A9) received the maximum score of 100% for actionability.

Table 6.4. Quality of the apps and the understandability and actionability of the information material

		MARS						PEMAT A/V	
ID	Name	A	B	C	D	Mean	E	U	AC
A1	Babble	3	3.5	3.7	3.8	3.5	14	75	33
A2	Birth & Beyond	2.2	4.8	2	4.2	3.3	10	100	100
A3	IFDC	3.4	4	3.3	3.5	3.6	14	75	100
A4	MyPreemie app	3.2	4.8	5	4.2	4.3	16	82	67
A5	My Prem Baby	3.2	3.8	3	4	3.5	14	55	100
A6	Neonatal Care	3	3.6	4	4.3	3.7	15	83	100
A7	Our Journey in NICU	1.8	3.3	1.7	2.3	2.3	8	75	100
A8	Preemie Mom Care	2.4	4	3.3	3.4	3.3	8	82	67
A9	PretermConnect	3.4	3.5	3	4.2	3.5	14	75	100
	Mean (SD)	2.8 (0.6)	3.9 (0.5)	3.2 (1.0)	3.8 (0.6)	3.4 (0.5)	12.6 (3.0)	78 (12)	85 (24)

MARS, Mobile App Rating Scale; PEMAT-A/V, Patient Education Materials Assessment Tool for Audiovisual Materials evaluation; A, Engagement; B, Functionality; C, Aesthetics; D, Information Quality; Mean, overall mean quality; E, subjective quality; U, understandability; AC, actionability.

Discussion

Principal results

This app review provides insight into the availability, content, quality of the apps, and the understandability and actionability of the information material for parents of preterm infants after hospital discharge. Nine apps were identified that provided information after hospital discharge, but the amount of information on the post-discharge period was limited in all apps. Only one app was of overall good quality, while the mean overall quality was between acceptable and good. The understandability and actionability of the apps were respectively moderate and good.

Although our inclusion criteria focused on the post-discharge period, the apps in this review contained primarily information for the NICU period. The lack of high quality and understandable apps found in this review is in contrast with the needs of parents of VPT infants after hospital discharge. Parents of VPT infants have reported challenges when they are at home regarding the availability and usability of information (8). For parents who struggle to seek information, finding an app with appropriate and reliable content will be even more difficult, particularly for those with low health literacy skills (29). Health care professionals, such as nurses, pediatricians or pediatric physical therapists have a responsibility to support parents in their search for relevant and reliable information during their hospital stay. As parental competence was found to decrease after discharge home, it is an important strategy to improve parental confidence in taking care of their VPT infant (30). When parents and infants are at home, without direct access to a health care professional, apps have the potential to provide health information to parents and can be accessed when and where needed.

Mobile apps can however not replace in-person care. Effective use of apps requires guidance from healthcare providers, as combining digital tools with professional support has been shown to enhance parental confidence (31). This is even more important for parents with limited health literacy or digital literacy, who are at higher risk of misunderstanding or misapplying information (18, 19). Our findings confirmed that the understandability of many apps is limited, largely due to complex medical terminology and text-heavy formats. This can particularly exclude parents with low health literacy, widening the existing digital divide (32). Improving understandability, for example through

audio, video, simplified language, and multilingual options, along with professional support, is essential to make apps usable and effective for all parents.

Apps that cover both the period in hospital and after discharge can be beneficial to parents by providing relevant information throughout the different phases. In a previous review of 18 apps in the NICU context (15), only five were included in our review, indicating that most NICU apps do not cover topics post-discharge. There was variability in the amount of post-discharge information, the topics, the emphasis within the topics, and how the information was presented. Unfortunately, the topics do not seem to correspond with the information needs of parents upon discharge (5, 8), such as daily infant care, neuromotor development, as well as the impact of prematurity on parents. Instead, most topics are focused on vaccinations, follow-up, and using home oxygen.

Despite the use of Dutch search terms, no Dutch apps were retrieved in the App stores that fulfilled the in- and exclusion criteria. The Dutch apps that were found in the Google search were no longer available in the Google Play Store or Apple Store. The majority of the apps evaluated in this review were last updated over one year ago. This lack of updates is in line with a scoping review about problems and barriers related to the use and implementation of apps (33) and, consequently, impedes usability and user experience which ultimately affects the effectiveness of applications. Apps without active maintenance quickly become outdated due to evolving technology, guidelines, and operating systems (36). This underscores the necessity of a viable business model and continuous refinement and maintenance after initial development (35). Sustainable funding for apps is essential but there are currently few resources available. Partnerships between industry and research may offer a possible solution for some apps.

Only one app had good overall quality, whereas the mean overall quality of the apps was merely acceptable. This is in agreement with an earlier app review, where the mean overall quality was also acceptable (15). Specifically, aspects within the domains of Engagement and Aesthetics could be improved. The apps scored particularly low on the engagement domain of the MARS, lower than acceptable. This subscale assesses whether the app is fun, interesting, customizable, interactive and well-targeted to the audience. Lack of engagement is a common barrier related to the use of mHealth apps and is associated with low adherence (33). Different functionalities can facilitate parental engagement with an app. A low rating on the Engagement domain suggests improvements are needed. Increasing engagement through entertainment appears not suitable for an app that provides information related to prematurity. However, the app

could be customizable or interactive, and should certainly be well-targeted to the audience. If not, this latter aspect would certainly hinder the use of the app. A positive finding from our review was that the domain 'information quality' of the apps was close to good. Reliable information is important as it may decrease parental stress, and supports better caregiving behavior (12). This also matches the parental needs for reliable information and is probably a result of the reputable sources (hospitals, universities) that developed the apps. This is in contrast with two previous studies in which only 31% and 40% of the websites provided accurate and reliable information for parents of premature babies (6, 11).

The understandability of the apps was scored as moderate, largely due to the primarily text-based information, indirect communication with users, and frequent use of medical terminology. In contrast, the app Birth and Beyond circumvented this problem by using only videos, in multiple languages. During stressful periods, such as hospital discharge, information should be presented in a clear and accessible manner, particularly for parents with low health literacy. For these individuals, the digital divide can be further exacerbated when the information is difficult to comprehend. The hospitals, universities and NGOs create apps with reliable information, but it may not be easily understood by all users. To meet the informational needs of all parents, apps need to be more understandable. Co-design that incorporates both healthcare professionals' and parents' perspectives can enhance app understandability by identifying the preferences and needs of the target group (36). Reducing text, written at accessible reading age levels, using multiple languages, and incorporating audio and visual formats may improve understandability.

Only two papers were retrieved that described the development of an app (27, 28), indicating a general lack of transparency about co-creation. None of the nine apps have been assessed for their impact on parental outcomes. A study on the NICU2HOME app (31, 37) showed that parental self-efficacy and satisfaction with care improved in parents of preterm infants. This mobile app has not been included in this review, as an access code was required. More research is needed to evaluate the use of apps, parental satisfaction and the effects of app use on parental outcomes.

Limitations

First, only English and Dutch apps, free and without an access code were included in the search, thereby possibly missing potentially relevant apps. Second, other

online resources that provide information to parents, such as websites, were also excluded. Also progressive web applications were not captured in our search, as these are not available in the searched app stores Third, the search for apps is time-dependent. Some apps are only available for a short time in the app stores, and replication of the search is therefore difficult. This became clear when apps identified through Google or PubMed were not available in the app stores. During the initial screening of app descriptions and features, followed by a secondary screening for inclusion, app content has been checked to decide whether it did also contain information related to the post-discharge period. Fourth, it may be possible that apps have been excluded during the initial screening because the description did not refer to information related to the post-discharge period. However, this information was then likely not substantial and would also not appeal to parents. Fifth, the assessment of the quality of the apps and the understandability and actionability of the information material has been done by the MARS and PEMAT A/V. These are validated tools used by professionals. The researchers were familiar with the parental needs for information (8) and did consider the parental perspective during the evaluation of the apps. However, direct information from parents of a preterm infant have not been taken into account. As parents are the key users, their experiences are most valuable and their engagement is important to ensure the content meets their needs. A next step would be to include parents to evaluate their experiences with good quality apps. Finally, although it was evident that information on the post-discharge period was limited, we did not quantify the amount of information provided in the apps.

Topics on post-discharge information were identified using a checklist, and compared to previously recognized parental needs for information. While the lack of information on the post-hospital discharge period was apparent, no specific measurement was conducted to assess the extent of information for the hospital or home environment. Furthermore, an assessment of the relevance of the topics was also lacking. Future work may establish new methods to incorporate these aspects.

Recommendations

During the post hospital-discharge period, parents of preterm infants need evidence based, reliable, and practical information. Mobile apps have the potential to offer this information in an accessible way. Currently, few good quality apps exist that contain reliable and understandable information, as the

My Preemie app or Preterm Connect. However, more relevant information that matches the needs of parents of VPT infants after hospital discharge is necessary. Future development of digital support tools should also consider solutions that bridge the gap between in-hospital and at-home care by extending access to apps currently limited to the NICU setting. Co-design with parents has been shown to improve the relevance and understandability of health apps (38).

We not only recommend that future apps should be developed or adapted in co-creation with end users but also that the development process is clearly reported. Research into the use and satisfaction of the parents should establish what information is key for parents, as well as how to deliver this information. In addition, the accessibility and understandability of an app needs to be evaluated among parents with a preterm born infant. The next step would be to evaluate the effect of the information app on parental outcomes as parenting skills, knowledge, and confidence

Conclusion

The current availability of mobile information apps for parents of preterm infants targeting the period after hospital discharge is limited and not in line with the high parental demand. Nine English apps were identified containing information on the post-discharge period. However, the apps contained limited content for the period at home. The overall quality of the apps was just acceptable, but the information quality was close to good. The understandability of the apps was moderate. Developing apps in co-creation with the end-users to better match their needs and increase the understandability are recommended.

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Chapter 7

An information app (e-TOP) to support parents' information needs during the first year at home after preterm birth: Development and usability study

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Abstract

Background: Parents of preterm infants often face challenges in transitioning from hospital to home, requiring reliable and accessible information to support their caregiving. Mobile health interventions have the potential to supplement post discharge education and empower parents by providing tailored, evidence-based information.

Objective: The primary objectives of this study were to develop an information app (e-TOP) using a participatory design approach, incorporating input from parents and health care professionals, and evaluate its usability.

Methods: A 2-phase, mixed methods design was used for this study. In Phase 1, the app was developed through iterative focus group discussions with parents of preterm infants, parents with limited health literacy, and TOP interventionists. In Phase 2, a usability study was conducted over 6 months with parents of preterm infants. Usability was assessed with a broad range of measurements: thinking aloud sessions, user engagement analytics, 2 questionnaires, the system usability scale (SUS), a customized satisfaction survey, open question sections, and semi structured interviews.

Results: The collaborative approach with end users and experts for the development led to a fully functional e-TOP app. Expert review and content validation ensured that information was clinically accurate, accessible, and relevant to parental needs. For the usability testing, a total of 58 families (116 participants) were recruited, and 69 participants actively used the app. The median cumulative e-TOP usage per participant in 6 months (26 weeks) was 39 minutes (IQR 8.8–53.0). The median number of actions was 64.0 (IQR 33.5–88.0). The e-TOP app received a median SUS score of 75 (IQR 67.5-80.0), indicating good usability. Participants rated their overall median satisfaction at 7.0 (IQR 7.0–8.0) out of 10. While the app was perceived as useful for finding information on prematurity-specific topics, engagement declined over time. Qualitative feedback highlighted a need for improved navigation (e.g., a search function), expanded content (e.g., motor development and sensory processing), and more interactive features (e.g., chat support and parental community forums).

Conclusions: The e-TOP app is a valuable digital resource that can supplement post discharge care by providing tailored, evidence-based, and accessible information for parents of preterm infants. While usability scores were high, engagement trends and retrieved feedback suggest the need for enhanced retention strategies, such as push notifications, timeline-based navigation structure, interactive tools, and adding (practical) content. Future iterations to improve the inclusivity of the e-TOP app require strategies to engage fathers and parents with limited health literacy skills.

Introduction

Background

In the Netherlands, 6.9% of the children are born prematurely (before 37 weeks of gestational age) (1). Although advances in perinatal and neonatal care have significantly improved survival rates for preterm infants (2), preterm birth still accounts for 66% of all deaths occurring before or during the first month after birth (3). Prematurity also has large adverse effects on children's motor, cognitive, and socioemotional development (4). Giving birth to a premature infant is also a disruptive start for parents (5). Parents of preterm infants often experience a range of mental health problems, such as anxiety and depression (6). The dependency on medical and health professionals can challenge the formation of a secure infant-parent relationship and parents' self-efficacy in their ability to independently care for their infant (7). The transition to home following discharge is known as challenging. Without the 24/7 access to health care professionals, parents of preterm infants feel unprepared for this new situation and lack confidence in their capacity to care for their infant (8). Despite efforts to improve parents' readiness for discharge and the availability of follow-up services, many parents of preterm infants continue to experience unmet needs for support and practical information after leaving the hospital (9, 10). As the infant's care needs and developmental milestones evolve during the first year, the informational needs of parents of preterm infants also shift accordingly. Providing tailored information about the consequences of prematurity and the specific health, behavioral, and developmental needs of premature infants can strengthen parental self-efficacy, enhance their sense of security in caring for their infant, and potentially reduce health care use (11, 12).

Search for information

After returning home, parents of preterm infants search the internet for information on topics, such as infant crying, choice of formula, common illnesses, and developmental stages. While they use search engines, social media, or mobile health (mHealth) apps, finding appropriate and reliable information on the unique challenges of caring for a preterm infant at home remains difficult (8, 9). The use of mHealth could support a shift from time- and professional-dependent access to information toward a more autonomous approach, empowering parents to obtain information whenever and wherever they need it. However, for mHealth apps to be useful and effective, they must address the specific information needs

of parents of preterm infants (13, 14). In addition, the app must be user-friendly (15) and the information accessible and easy to understand for parents across varying levels of education and health literacy (16).

mHealth apps

mHealth education interventions for parents of premature infants were developed over the last decades. However, Richardson's evaluation of mobile apps targeting parents of infants in the neonatal intensive care unit (NICU) revealed that many apps are lacking quality and credibility or do not contain content identified by parents as important (17). In contrast, the study on the NICU2HOME app provides high-quality information on infant and self-care during and after the NICU stay and showed promising results in enhancing parental self-efficacy and discharge preparedness (18). Despite the rich body of literature regarding parental needs for information after discharge, available apps targeting the post discharge period are limited. A systematic app review of mobile information apps for parents of preterm infants after hospital discharge found that, while the overall quality of the identified apps was acceptable, they insufficiently addressed key post discharge concerns, such as information on transitions in nutrition, sleep patterns, or developmental stages (19). This lack of targeted content may contribute to the limited usage and adoption of these apps, a common pitfall when end users and stakeholders are not adequately involved in the design process (20). Participatory design approaches, such as the design thinking method, ensure that user requirements are accurately captured from the perspective of the end users, thereby enhancing usability and facilitating effective implementation (21-24).

The Dutch context: TOP program

In the Netherlands, a 1-year home-based responsive parenting intervention (TOP program) is standard care for very preterm infants and their parents and is currently expanded for moderate to late preterm infants. Pediatric physical therapists with an additional 1-year TOP training and affiliated with the Dutch Expertise Center for Premature Infants (EOP) carry out the TOP program. The strength-based and process-oriented intervention consists of 7 key strategies to enhance parental understanding of the infants' behavioral and developmental needs (25). An important key strategy is to provide parents with information about their infant's behavioral and developmental needs. To enhance parents' knowledge and understanding of their infants' needs, TOP interventionists

provide verbal information transfer and an individualized written parental report with strength-based recommendations accompanied by photos taken during the home visit. Despite the benefits of face-to-face care, there is a need for a complementary mHealth intervention that enhances information retention, supports parental confidence in managing their infant's care, and potentially reduces their reliance on health care professionals (9, 14). Therefore, we aim to complement and enhance the existing Dutch TOP program by addressing gaps in information retention and supporting professionals in delivering reliable information to parents. Before conducting more extensive research to measure its effectiveness, developing an information app that ensures that user requirements are effectively incorporated and is tailored to the specific needs of parents of preterm infants is crucial.

The aim of this study was two-fold: 1) to develop a post discharge information app (e-TOP) for parents of premature infants; and 2) to evaluate its usability.

Phase 1: Development of the e-TOP app

Phase 1 Methods

Setting The study was performed between October 2021 and November 2023 by the EOP, affiliated with the Amsterdam University Medical Center, in collaboration with the Amsterdam University of Applied Sciences.

Ethical considerations

The study was approved by the Medical Ethical Committee of the Stichting Amsterdam UMC (NL78996.018.21). All participants received an information letter detailing the study's objectives, timeline, content, target group, data management practices, and privacy considerations. Subsequently, informed consent forms were obtained. Participants were officially enrolled in the study upon providing written consent. Data were pseudonymized. Participants did not receive any compensation.

Participants

The user-centered design approach for the development of the prototype started with 3 focus groups, 1 with TOP interventionists and 2 with parents of preterm infants. A pragmatic sampling approach was used, drawing on available data from the EOP center to identify and invite participants (TOP interventionists) with relevant experience across different contexts (26). For focus group 1, TOP

interventionists with more than 5 years of experience, representing different geographic and socioeconomic regions, were approached. For the parent focus groups, participating TOP interventionists approached potential eligible and interested families within their caseload. Parents were included if they 1) participated in the TOP program for at least 3 months and 2) were able to understand and speak the Dutch language. Focus group 3 specifically involved parents of preterm infants with limited health literacy (LHL). These parents were identified and recruited by the TOP interventionist by using the following 2 additional criteria: 1) parents struggle with understanding and applying the information provided during the intervention or in the parent report and 2) fulfill the criteria for LHL according to the Dutch checklist to recognize LHL (27).

Focus group meetings

The script for the focus group meetings was developed collaboratively with the research team, guided by the 2 main objectives of the sessions (Appendix 7.1). First, to verify the identified need for information on the 10 topics previously found in our qualitative research (13) and to enrich these with participants' personal experiences. Second, to elicit preferences regarding content, topics, design, and navigation of the app.

To address the first objective, the research team formulated open-ended questions and prompts for each of the 10 topics. These were designed to stimulate reflection, encourage sharing of personal experiences, and allow for the addition of new themes. Examples and short scenarios were incorporated to help participants relate the topics to their own context.

For the second objective, the script included guided activities involving visual and textual materials, such as screenshots of existing parenting apps, information cards, mock-ups, and text samples. These were selected and designed by the research team to support discussion on specific design features (e.g., tone of voice, level of detail, and visual presentation) and to facilitate comparison between different styles. See Figure 7.1; information cards and Figure 7.2; mock-ups.

The draft script was reviewed in 2 rounds by the research team to ensure alignment with the study objectives, clarity of language, and feasibility within the planned 2-hour sessions. Minor adjustments were made following pilot testing with a small group of colleagues to refine the sequence of questions and ensure smooth transitions between topics. We conducted 3 focus group sessions. In the first meeting, 8 female TOP interventionists participated. Five

interventionists worked in an urban setting with a diverse population in a challenging socioeconomic environment.

In the second focus group, 9 parents (5 mothers and 4 fathers) participated. Their mean age was 35.6 years (SD 5.8). For 5 parents, it was their firstborn, and for the other 4 parents, it was their second child. Their infants' mean gestational age was 30 weeks and 2 days (SD 19.0). In the third focus group meeting, 6 mothers with LHL participated with a mean age of 31.6 years (SD 2.2). For 5 mothers, it was their firstborn infant, and for 1 mother, her third child. Their infants' mean gestational age was 29 weeks and 1 day (SD 21.8).

Figure 7.1. Information cards.

Figure 7.2. Mock-up.

The three 2-hour online focus group meetings were organized in the evenings, which was indicated by participants as the most convenient time. The focus groups were facilitated by an independent female researcher, MM. She was not involved in the study design or analysis. The first author (MF) attended the focus groups to take field notes but did not lead the discussions. The 3 focus group meetings were audio-recorded, relistened to, and summarized. Findings were discussed within the research team.

Phase 1 Results

Overview

The results of the focus group discussions with TOP interventionists and parents provided valuable insights that shaped the development of the e-TOP app, directly influencing its content and design refinements. TOP interventionists recommended not to use the age-based developmental norms strictly, as individual variation among preterm infants is substantial. Instead of providing medical guidance, the app should direct parents to contact their general practitioner or pediatrician when necessary. Additional topics identified by participants were the digestive system, reflux, constipation, medication management, sensory overload and stimulation, parent-infant attachment, and bonding. Parents expressed a preference for clear, supportive, and actionable information. Information should have a layered content structure, allowing simplified messages and scientifically substantiated information. A summary of these findings is provided in Table 7.1. We used them as a guideline for the further development of the e-TOP app.

From prototype to a functional e-TOP app

To create content for the app, experts were recruited through the researchers' network. Health care experts were selected based on their expertise regarding prematurity or related topics, such as feeding, sleep, or motor development. To address user needs, the following 4 steps were taken. First, the topics parents indicated in our prior research (13)were complemented with the findings of the focus group meetings. Experts drafted comprehensive information on these topics. Two members of the research team (MF and EM) finalized the content in 3 rounds of review and refinement. Second, the Dutch Expertise Center for reducing health inequities provided support to improve the readability, whereafter the text was simplified by the first 2 authors (MF and EM) to that of the

Table 7.1. Results of focus group meetings

Topics	Focus group 1[a]	Focus group 2[b]	Focus group 3[c]
Associations participants have with a digital information source.	Reliable and user-friendly digital source aligns well with the needs of young parents.	An app can offer practical guidance supporting in everyday care. Given the limited experience of general practitioners and Dutch health services with prematurity, the app could also be a valuable source.	Participants expressed curiosity and recognized the usefulness of an app in addressing their concerns
What questions do parents have?	Ten topics most frequently searched align with participants concerns, validating the app's content. In addition, practical information on adequate sensory input and use of slings is needed.	Different experiences with gavage feeding at home, a need for more tailored information. What can I do with my premature infant?	Participants sought information on baby positioning, especially with preferred head position. In addition to the showed topics: reflux and gavage feeding tips. Information about constipation.
About which topics do parents seek additional information?	Information about constipation, intestinal cramps, reflux, or medication.	Feeding, sleep patterns, sensory overexcitability	Inconsolable crying. Psychological support for anxiety. Premature clothes. Information about long term effects of oxygen.
Which support do parents need?	Finding psychological support for themselves.	Need for a readily accessible professional after discharge.	Information on available services, particularly for single moms.

Table 7.1 continues on next page.

Table 7.1. ***Continued***

Topics	Focus group 1[a]	Focus group 2[b]	Focus group 3[c]
Where do they look for information?	Online communities including Facebook group and consumer association websites.	Parents shared their favorite websites but they are often not specific for premature infants.	Online forums such as baby 24 are used. Other parents with experiences serve as key sources of information.
Where do they look for information?	Online communities including Facebook group and consumer association websites.	Parents shared their favorite websites but they are often not specific for premature infants.	Online forums such as baby 24 are used. Other parents with experiences serve as key sources of information.
Presentation of information	Adaptability to literacy levels. Videos and animations in multiple languages for less literate parents.	Varied content preferences. Different preferences, some participants liked text-based information, others favored videos.	Preference for animation (visual oriented). Practical demonstrations with videos for e.g., making the crib. Interactive features like chat box for quick response. Peer-experiences. Some valued real-life success stories, others were not interested in fellow sufferers.
Navigation	A structured topic design, with a FAQ chapter.	Search button to improve content discoverability.	Clicking through for more detailed information.

Topics	Focus group 1[a]	Focus group 2[b]	Focus group 3[c]
Information cards	Reassuring and supportive tone is essential. Normalizing and comforting tone of voice is important.	Concise messages can raise questions. The messages should not oversimplify.	Informative, necessity to be able to search further for more extensive information.
Text validation	Be aware of tone of voice, clarity of the information.	Comforting information is important. Informal tone of voice is nice, it related to parents and feels personal.	Easy-to-read text, supplemented by practical examples on what to do. Information can be brought more casual, like you talk to a trusted friend or relative. It needs to be valid information.
Depth of information	Layered information structure. Parents need to be able to explore in-depth details or stay at a simpler level.	Different headings/titles can help to make content more accessible.	Information should clearly explain differences in care for premature infants.
Scope of information	Coverage for the first year, aligning with the TOP program.	Guidance on more complex cases, should refer parents to the health care professionals. Potential integration with a hospital app could enhance usability.	Developmental milestones per week/ updates about what your baby is doing in relation to other infants (corrected for prematurity)

[a] TOP interventionists. [b] PT parents. [c] PT parents with LHL.

reading ability to understand everyday health information (B1/B2). Key messages for each topic were formulated at level A2 for parents who prefer to read short sentences with familiar words (28). In addition to text, a read-out-loud function was added. Third, the first 2 authors (MF and EM) included images and picture stories. Fourth, video content, either developed by health care experts from different fields or retrieved from the EOP library, was parent-infant interaction, and explanations of infant behavioral signs. To clarify the content, videos had voice-overs and subtitles. Simultaneously with creating content, we collaborated with a design company (ZIGT) to build a functional prototype of a web app accessible on smartphones, tablets, and desktop devices. The research team organized and posted all content leading to the final functional e-TOP app (see Figure 7.3). added. Videos consisted of interviews, demonstrations of e-TOP app.

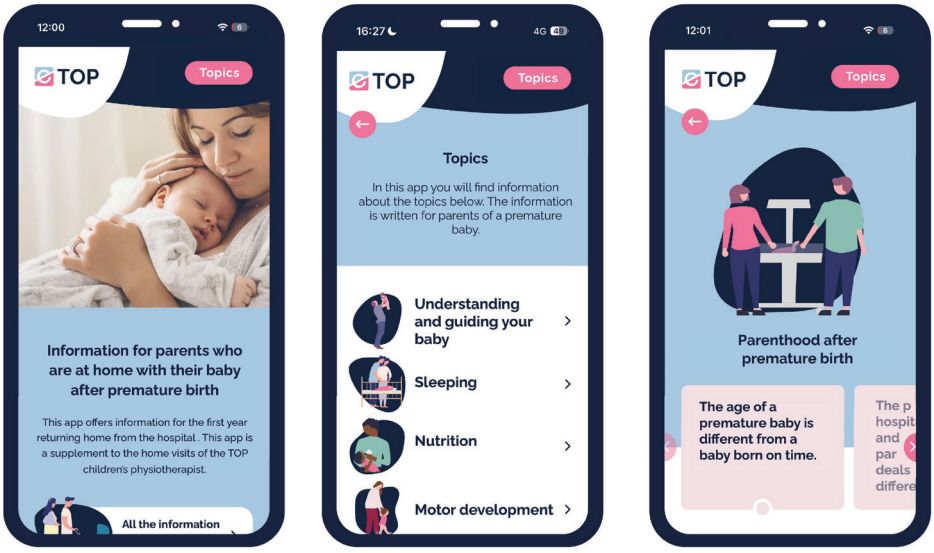

Figure 7.3. e-TOP app.

Phase 2: Evaluation

Participants

For the evaluation of the e-TOP app, the sample was composed of parents with a very preterm born infant (below $32^{0/7}$ gestation) and moderate preterm born infants ($32^{0/7}$–$34^{6/7}$ weeks of gestation). Parents of very preterm infants received the TOP program (routine care, reimbursed by the basic package of insurers), and parents of moderate preterm infants received a shortened TOP program in

a study context (29). In addition to the home visits by the TOP interventionists, all participants received access to the e-TOP app.

Procedures

TOP interventionists were recruited using an invitation email distributed by the EOP center. Fifteen TOP interventionists responded to be available for participation in the study. They received a 1-day training in the intervention and research protocol including the informed consent procedure. The enrollment took place between November 2022 and March 2023. TOP interventionists introduced the study to eligible parents within their caseload. The requirement for parents to take part in the study was to be able to read and speak Dutch. When interested, parents received an information letter from the study center. If they agreed to participate, an informed consent visit was scheduled. Upon signing the informed consent, both parents received a unique access code and an animation video explaining how to install the e-TOP app. In addition, at the start (T0), parents were invited via email to complete a questionnaire on sociodemographic information including age, educational level, employment status, and family structure. Parents had continuous access to the app for the duration of 6 months. During this period, Matomo analytics was used to collect user data and thinking aloud tests were performed. At the end of this period (T1), parents received a follow-up invitation to complete questionnaires assessing their experiences, and 10 participants were invited for an interview.

Usability

For a comprehensive evaluation of the usability of the app, we used qualitative and quantitative measurements. User data provided insights into objective usage engagement, while qualitative data highlighted usability needs and identified issues for further improvement (30).

Usability testing: Thinking aloud

Thinking aloud tests were used to gain insights into the ease of use and navigation experiences with the app (31, 32). While navigating the app, parents were encouraged to express their thoughts, frustrations, questions, and feedback in real time. The tests were conducted by 2 students from the faculty of health of the University of Applied Sciences Amsterdam, supervised by author VV. For the sample technique, a stepwise approach was used to ensure that the final participant list for the thinking aloud session included a varied sample across

different levels of app engagement. The selection process involved the following steps: 1) all families were initially screened for e-TOP app installation. 2) For each participant, the total app usage time (in seconds) was calculated. 3) Quartiles of app usage time were determined, and participants were divided into 4 usage groups. 4) Each group was randomly ordered using a randomization tool. 5) Three participants per group (n = 12) were approached by email for participation. Participants were asked to complete 3 tasks: 1) determine when your infant can pick up a small object; 2) identify what actions you can take as a parent when your child is anxious about touch; and 3) calculate your infant's corrected age. After completing each task, participants rated the difficulty using the Single Ease Question (33), which scores task performance on a scale from 1 (very difficult) to 7 (very easy). Afterward, parents were asked questions regarding the content (e.g., "Are there any topics you would like to add?"), usability (e.g., "How was your experience with the e-TOP app?"), and comprehension (e.g., "Did you understand the answers you found when searching for information?"). These sessions were screen-recorded, and audio transcripts were generated.

User data

User data were monitored and analyzed with the web analytics tool Matomo (34). Users were defined as those who accessed the e-TOP app more than once. The main metrics of interest were installs, visits, visit duration and change of usage over time, and visited topics.

To allow participants to provide immediate feedback on the app, the e-TOP app included a prompt at the end of each page asking, "Was this information helpful?" with 2 response options: yes or no. After selecting an option, a comment box appeared, allowing users to provide additional feedback.

Usability questionnaires

The Dutch version of the system usability scale (SUS) was used (35). The SUS is an easy-to-use, valid, and reliable questionnaire for usability assessment of mHealth apps. The scale consists of 10 statements, which can be completed within 10 minutes. All items are scored on a 5-point Likert scale from "strongly disagree" to "strongly agree." Scores per item are converted to an overall SUS score, which ranges from 0 to 100, with higher scores indicating better usability. Overall scores from 0 to 50 indicate "not acceptable," scores from 51 to 67 indicate a marginal level of usability, and scores from 68 to 100 indicate "acceptable to excellent" levels of usability (36).

To measure user experience and appropriateness of the content, a customized 10-item questionnaire was used. Seven statements were scored on a 5-point Likert scale from "strongly disagree" to "strongly agree." Two statement examples are: "The e-TOP app helps me to find information about premature birth" and "The pictures and videos help me to understand the information." Overall satisfaction with the app was scored on a numeric scale from 1 to 10 with 1 indicating very dissatisfied to 10 indicating extremely satisfied. Two open-response questions allowed participants to provide unrestricted and more subjective answers: "What information did you miss when using the e-TOP app?" and "Do you have suggestions to improve the e-TOP app?"

Interviews

The interview guide was developed in collaboration with the research team. Questions were formulated to address predefined aspects considered critical for evaluating the app's usability. These aspects were derived from the study objectives and focused on identifying strengths, limitations, and opportunities for improvement. To gain a more in-depth understanding of the experiences with the e-TOP app, a subgroup of participants (n = 10) was invited for a semi structured interview with the first author (MF) to discuss their experiences with the e-TOP app in more detail. Purposeful sampling, including educational level, gender, and e-TOP app usage, was used to obtain a heterogeneous sample. Interviews were scheduled at a time convenient for the parents (by phone or video call). The interview guide was set up to elicit direct, actionable feedback on app usage (Appendix 7.2).

Data analysis

Questionnaire data were collected using the Castor database (Electronic Data Capture, Ciwit BV, 2021). Descriptive statistics were used to describe the participants and the user data (installs, visits, visit duration and change of usage over time, and visited topics). Questionnaire data were checked for normal distributions using a graphical summary of data and descriptive statistics. The median score with IQR was used in case of skewed distributions.

For the qualitative data collected through thinking aloud sessions and interviews, a pragmatic thematic analysis approach was used (37). For the thinking aloud analysis, initial open coding was analyzed and discussed within the project group and subsequently categorized into themes. Subthemes were then developed under each main theme to capture more specific insights. In

addition, user comments and open questions from the questionnaires were evaluated and categorized by the first author (MF). Ultimately, the qualitative analysis resulted in 4 overarching themes: user experience, content, navigation, and functionality.

Phase 2 Results participants

A total of 69 families with a preterm infant were assessed for eligibility by 15 TOP interventionists. Of these, 58 eligible families, consisting of 58 fathers and 58 mothers, agreed receiving the first questionnaires. Five families (10 participants) withdrew from the study after signing informed consent and receiving the first questionnaires.

At T0, 61 participants to participate, signed informed consent, and received the completed the demographic questionnaire and access code for the e-TOP app (see Figure 7.4 for flow diagram). For participant characteristics, see Table 7.2.

Table 7.2. Participant characteristics

Participant characteristics	N (%)
Parent	
Mother	42 (69%)
Father	19 (31%)
Family status	
Firstborn child	37 (61%)
Family status of 2 parents	61 (100%)
Language	
Dutch language for health care information	60 (98 %)
Education[a]	
Lower	2 (3%)
Intermediate	22 (36%)
High	37 (61%)
Employment	
Fulltime	35 (57%)
Part time	25 (41%)
No employment	1 (2%)

[a] Highest educational level completed Low level education refers to primary school, vocational education, lower of middle general secondary education. Intermediate: refers to higher secondary general education, pre-university education. High; Higher vocational or university.

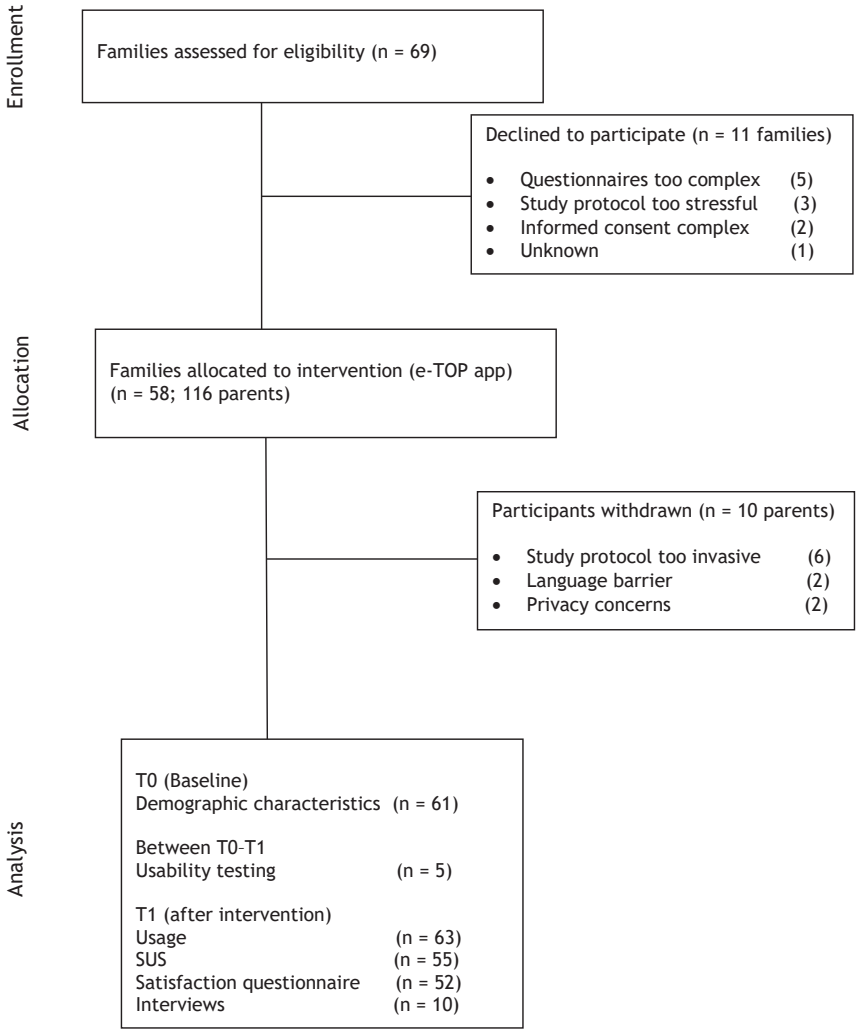

Figure 7.4. Flow of participants.

Usability results

Thinking aloud

Ultimately 5 participants (3 fathers and 2 mothers) were willing to schedule an appointment to perform the Thinking aloud tasks. The ease of use was rated as follows: Task 1 had a mean score of 6.4 (SD 0.55), Task 2 a mean of 6.0 (SD 0.89), and Task 3 a mean of 6.0 (SD 1.22) on a scale from 1 to 7, indicating that all 3 tasks were easy to perform. Participants appreciated the app, the tone of voice, and the understandability of the information. Participants provided explicit and

actionable recommendations regarding content and functionality. For example, regarding content, participants expressed a desire for more information on health issues and medication. They also expressed interest in instructional videos on techniques, such as holding and carrying their baby. Regarding functionality, a home button, a scroll arrow, and a search function were missed.

User data

In total, 69 participants (47 mothers and 22 fathers) used the personal access code to log in. Due to ad blockers, tracking data were available for 63 participants. The median cumulative e-TOP usage per participant during 6 months (26 weeks) was 39 minutes (IQR 8.8–53.0). The median number of actions was 64.0 (IQR 33.5–88.0). Most actions and the largest time spent on the app were registered during the 2 weeks after receiving the access code. See Figures 7.5 and 7.6 for further details. Multiple participants visited all 10 topics during the first 2 weeks. The total amount of visits per topic declined after this period with another peak in visits between weeks 11 and 17. The most visited topics in the app were nutrition, sleep, and correcting age for prematurity (Table 7.3).

Table 7.3. Visited topics on the e-TOP app

Topic	Total number of visits	Peak visits (week)
Nutrition	81	17
Sleep	79	6
Correcting age	54	3
Understanding your baby	49	4
Long term consequences	44	14
Motor development	42	11
Returning to work	33	4
Parenting	25	4
Infant health	23	4
Follow-up	21	9

Usability questionnaires

A total of 55 participants completed the SUS, yielding a median total SUS score of 75.0 (IQR 67.5–80.0), indicating generally good usability. The app was rated positively for its ease of use, functional integration, and user confidence.

Participants gave the app an overall satisfaction of 7.0 (IQR 7.0–8.0), on a scale of 1–10. Content satisfaction was rated with a mean score of 3.7 (SD 0.89)

and the app's appearance was considered attractive, with a mean score of 3.6 (SD 0.83) on a scale from 1 to 5. Participants found the pictures and videos particularly helpful (mean 4.0, SD 0.56) and expressed that the app was helpful in addition to face-to-face care from the TOP interventionist (mean 3.5, SD 0.99).

Qualitative data

Data collected through thinking aloud sessions, interviews, and written comments from the app's comment box were categorized in 4 themes: user experience, content, navigation, and functionality (see Table 7.4).

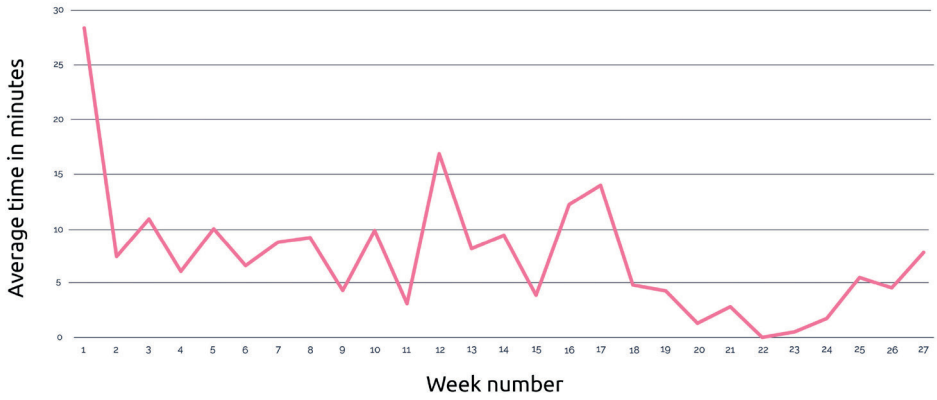

Figure 7.5. Time spent per week.

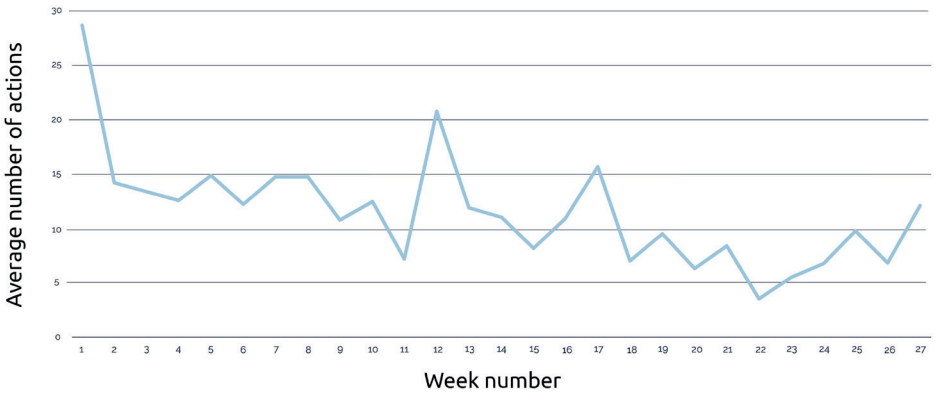

Figure 7.6. Actions per week.

Table 7.4. Integrated results matrix for quantitative and qualitative data

Themes	Quantitative findings (questionnaires and Thinking aloud)	Qualitative findings (interviews and comment box)	Meta-inference
User experience	Overall satisfaction: Median 7.0/10 (IQR 7–8) Visual attractiveness: Mean 3.6/5 (SD 0.93)	Users appreciated the depth and specificity of information and the app's tone. However, they missed tailored responses and desired more interactive features.	Parents appreciate the user experience, yet desire more individualized and interactive features.
Content	Content rating: Mean 3.7/5 (SD 0.89) Visual helpfulness: Mean 8.0/10 (SD 0.56)	Content was viewed as highly relevant and unique to prematurity. Suggestions included more on motor development, practical care, and instructional videos.	Content meets core needs, but parents want more concrete, in-depth, and visually supported information tailored to daily care.
Navigation	Median total SUS[a] score of 75.0 (IQR 67.5–80.0)	Parents found the app easy to use and appreciated its tone; they missed a search button and clearer navigation.	Usability is high, but navigation challenges (especially in locating specific info) call for interface redesign and better structure.
Functionality	Task ease thinking aloud: Mean: 6.1/7 (SD 0.89) Usage: Median 39 min. (IQR 8.8–53) over 6 months	Parents appreciated the functions in the app, but had some suggestions for improvements.	Functionality is strong but could be elevated by improved orientation, content sharing, and age-specific organization (e.g., timeline).

[a] SUS: system usability scale.

User experience

Participants appreciated the specificity and depth of information provided yet expressed concerns about retrieving tailored answers to specific situations. As a participant in the interview stated, "The only thing I couldn't find was whether it's possible to swim with a premature baby and at what age that is safe". The app facilitated users' understanding of infant behavioral cues and sleep patterns. However, parents also preferred direct interaction with a TOP interventionist, indicating a desire for more personalized and human support. For example, a mother stated in the interview that "The TOP interventionist explained it to me personally, so I found that easier and more comfortable."

Suggestions were given to incorporate interactive features, such as chat support or a parent community, indicating a preference for increased interactivity and social connection within the app.

Content

The content was generally deemed useful and relevant to the specific needs of parents with premature infants. As 1 participant noted in the comment box, "It gives information you cannot find on the internet, all content is related to premature birth." Suggestions for improvement emerged from both the interviews and the comment box. Requests for additional practical tips, along with more in-depth information on motor development and health-related concerns, highlighted opportunities to expand the scope of information. As a participant stated during the interview: "More information about motor development, like crawling, creeping, and sitting, would be helpful." Another parent expressed during the interview, "I would like there to be more concrete information, such as a chart with a baby weighing this much should drink this much." Videos were highlighted as particularly effective for conveying complex concepts, such as sleep patterns and infant signals.

Navigation

While users found the landing pages clear and well-organized, they reported difficulties with lengthy chapters and locating specific information. As a participant during the interview reported: "Searching for specific information is more difficult if you don't know under which chapter to search." Another participant reported in the comment box: "The sub-chapters contain too much information and not so easy to find answers to your questions." The need for enhanced navigation design, such as the inclusion of a search function, a home button, and scroll arrows, was suggested and could enhance information retrieval. A participant explained during the thinking aloud session: "If there was an arrow next to the app indicating that there's more room to scroll down, you might have looked further."

Functionality

A timeline-based structure reflecting the developmental stages of premature infants was suggested as a potential solution to improve content organization. One participant stated in the comment box: "It would also be helpful if there was a timeline showing what a child should do at each (corrected) age." The

ability to share information easily with family members, particularly regarding hygiene practices for visits, emerged as a key functional improvement.

Discussion

Overview

This study aimed to develop and evaluate a digital information source (e-TOP) to address the specific information needs of parents caring for preterm infants during the first-year post discharge.

Development of the app

To develop the e-TOP app, a user-centered design approach was used. The feedback from the 3 focus groups with TOP interventionists and parents of premature infants, including parents with LHL, directly influenced the app's content and design. The cocreation and iterative process between experts, the design company, and the research team was highly successful in shaping the app's functionality and refining its design and content. This collaborative approach ensured that the final prototype of the e-TOP app was both user-friendly and rich in relevant, evidence-based information, tailored to meet the diverse needs of its users. This methodology aligns with established practices in mHealth app development, emphasizing the importance of participatory design including health professionals to meet user needs (21-24).

Parents with LHL were actively engaged during the focus group meetings in the developmental phase of the e-TOP app. They expressed a need for clear, actionable information, preferably content in multiple formats, with an accessible tone and simplified messages. The app therefore incorporated features, such as simplified language (Level 1), multimedia content, and "read-out-loud" functionality to accommodate parents with LHL, with videos and text at different reading levels. This preference is supported by research indicating that visual-based interventions, particularly videos, are effective in improving health literacy and comprehension of health-related information (38).

Usage and usability

Participants engaged with the app for a median total of 39 minutes (IQR 8.8–53.0) during the 6-month study period. The number of recorded actions per participant had a median of 64.0 (IQR 33.5–88.0), but usage varied significantly between participants. Differences in purpose, target groups, and the lack of user engage-

ment metrics make comparison to other parenting apps challenging, as also described in Li's systematic review of commercial mHealth parenting apps (39). The e-TOP app is designed as an additional information source; if we compare the results to educational health apps, longer but less frequent sessions are found (40). This found gap calls for established standardized benchmarks for digital engagement in neonatal mHealth interventions.

While the app was perceived as useful for finding information on prematurity-specific topics, user data revealed a decline in engagement after the initial 2 weeks. These differences in intent to use versus lower non usage attrition are found in other web-based parenting interventions (15). The e-TOP engagement peaked at week 11 and week 16. Parents revisited the app at key developmental milestones, such as rolling over, reaching for toys, and the transition to solid foods. Integrating features such as search functionality, interactive (push notifications and chat function), and timeline-based navigation could enhance the e-TOP app's long-term utility (41, 42).

SUS distribution with a mean SUS score of 68 is considered a suitable usability for Digital Health apps (43). The e-TOP app demonstrated acceptable-to-good usability with a SUS score of 75.0 (IQR 67.5–80.0). By complementing the SUS with a customized satisfaction survey and data collected through thinking-aloud sessions, interviews, and written comments from the app's comment box, this study followed best practices of combining quantitative ratings with rich qualitative findings for the usability testing (30). Participants provided actionable feedback across key areas. By integrating these improvements, the e-TOP app can enhance usability, ensuring it remains a valuable long-term resource for parents of preterm infants.

Limitations and opportunities for future research

The total study period of 2 years and restricted finances necessitated pragmatic approaches to the evaluation of the usability. We used several qualitative and quantitative methods, but we had to make compromises impacting the study methodology, such as the reliance on a single author for qualitative data analysis which may introduce bias. Integrating multiple researchers into the thematic analysis process not only enriches the analysis through diverse perspectives but also enhances the credibility and trustworthiness of the research findings (44).

Although parents with LHL contributed to the development of the app, the study faced challenges in recruiting parents with LHL for the evaluation phase. Their limited representation in the evaluation phase underscores a broader

challenge in engaging these parents in research (45). Barriers such as complex consent processes and digital questionnaires have hindered participation in our study. Allowing parents to first establish a connection with the researchers or interventionists and using postponed consent is expected to promote inclusivity. Also, verbal consent or simplified forms could foster inclusivity. Although improved comprehension and trust may have positive effects on study enrollment, we emphasize the active involvement of the target group in the early stages of research projects when developing the study design, procedures, and the selection of measurements (46).

This study had a high attrition of the fathers; only 32% of the fathers used the app and 25% completed the usability questionnaires. In general, retaining participants in studies using mHealth apps has been described as challenging (47). The finding of higher attrition among fathers aligns with the results of the study on the NICU2HOME app (Garfield (18)), in which high usability scores were similarly observed among mothers, but lower engagement among fathers was noted (10). Xie (48) also found in her systematic review of digital interventions for fathers of infants from conception to 12 months postpartum that fathers have expressed the need and desire for access to relevant, accurate, and up-to-date information on infant care and challenges associated with new parenthood and called for interventions that target father identified priorities (49). These studies and our findings highlight the importance of gaining a deeper understanding of perceived barriers to engaging fathers.

The usability evaluation confirms that the app was responsive to the needs of parents with a premature infant. Given the evidence supporting user-centered iterative development, we strongly advocate for proceeding with the implementation of the e-TOP app, ensuring it remains a valuable and available resource while integrating continuous user feedback by using real-time analytics, for example, engagement metrics (50, 51). Future research should focus on assessing the effectiveness by evaluating how app usage translates into improved parental confidence and decision making in caregiving with the goal of reducing dependency on health care professionals and fostering parental autonomy.

Conclusion

The e-TOP app has demonstrated strong potential to complement information transfer to the face-to-face care after discharge and to bridge the information gap for parents when they leave the hospital. While usability findings are promising, continued development, customization, and integration of the

app with health care systems are necessary to maximize its long-term impact. By addressing user engagement, content expansion, accessibility, and health care integration, future empowering parents of preterm infants and improving versions of e-TOP can become an essential tool for neonatal post discharge care.

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Appendix 1: Focus group outline and key questions

This appendix outlines the structure and key discussion points used during the focus groups. The sessions were moderated using examples and visual aids to facilitate meaningful conversations.

1. Opening question

What are your first associations when you hear about an online information source (e.g., an app) for parents of premature infants?

2. Content-related questions

a. Practical concerns shared by other parents
- Do you recognize the following concerns raised by other parents?
- Why does my baby groan so much during sleep?
- Until what age should I correct for prematurity?
- What topics have you personally searched for online?

b. Examples from other parents
- “My infant has had a cold for a few days—do I need to consult a doctor?”
- “What can I expect when my child starts school?”
- “Is mental health support available for parents?”
- “What kind of support did you need after returning home from the hospital?”
- Some parents mentioned struggling to build confidence in their parenting role or wished they had received more practical guidance.

c. Sources of information
- Where do you typically find information?
- Other parents reported seeking advice from healthcare professionals, family, friends, online searches, or Facebook groups.

3. App development questions

a. Information structure preferences
- Do you prefer information organized by topic or by frequently asked questions? (Mockups were used during this discussion.)

b. Feedback on mockups
- Are the information cards and texts shown in the mockups clear and sufficient for your needs?
- Different styles of mockups were presented (text-only, images, and combinations).

4. Validation of text content

Participants were shown or read sample texts and asked to respond to the following:

a. Open feedback
- What are your impressions of the text you just read?

b. Rating statements
Participants rated the text using a 4-point Likert scale (1 = strongly disagree, 4 = strongly agree) for the following statements:
- The text feels personal.
- The text is reassuring.
- The text is alarming.
- The text is informative.

c. Tone of voice
- What tone of voice do you prefer for such content?
  (Examples were given: healthcare professional, grandmother, peer/buddy.)

5. Preferences for visual content

a. Use of visual aids
- What are your preferences for visual content (e.g., animations, videos, pictorial stories)?
  (Examples were presented to the participants to facilitate this discussion.)

Appendix 7.2: Interview guide for evaluating the e-TOP app

1. General use and engagement
- Did you use the e-TOP app?
- Could you explain the reasons for using or not using the app?
- How would you describe your overall experience with the e-TOP app?
- Did you use the app during specific moments or situations?

2. Specific information needs
- What type of information were you looking for in the app?
- Did the app provide the information you were seeking?
- Can you provide an example of a time when you could not find the information you were looking for?

3. Content and accessibility
- How did you experience the content provided in the app?
- What were your impressions of the way information was presented (e.g., text, videos)?
- Could you elaborate on whether the level of detail and depth of information was sufficient?
- Were you able to navigate through the chapters to find relevant information?
- Did you find the absence of a search function to be a limitation?

4. Alternatives and additional needs
- What did you do when the information you sought was not available in the app?
- Did the videos help you to better understand the content?
- Could you relate to the people or situations shown in the videos?
- What suggestions do you have to improve the app to better meet your information needs?

Chapter 8

Summary of findings and general discussion

The overarching aim of this thesis is to identify, develop and evaluate strategies to the impact of the post-discharge responsive parenting program for prematurely born infants and their parents (TOP program). To achieve this goal, the thesis focuses on three key areas for improvement: 1) strengthening the quality of TOP program implementation; 2) expanding the program's reach to a broader target population; and 3) refining and adapting intervention strategies. This chapter summarizes the main findings of all studies, followed by an in-depth general discussion and a broader perspective on implications for future research, policy, and clinical practice.

Summary of findings

Chapter 2

This study developed and validated the Fidelity of Implementation Tool for the TOP program (FITT), a responsive parenting intervention for very preterm (VP) infants (born < 32 weeks gestational age (GA) and/or < 1500 g). Three consecutive phases were used following an iterative and co-creative process. Twenty intervention videos from nineteen families were used to evaluate the psychometric properties. Results showed strong interrater reliability and good internal consistency across subscales, indicating that the FITT can be applied consistently by different evaluators. In addition, the tool demonstrated good discriminative ability, effectively capturing meaningful variation in provider performance. The final FITT, consisting of adherence and competence subscales plus a general impression score, provides a reliable, feasible, and clinically useful tool to measure fidelity in the execution of the TOP program.

Chapter 3

This feasibility study evaluated the applicability of the modified TOP program, originally developed for very preterm infants, to a new target group of moderate preterm (MP) infants (32–35 weeks GA, > 1500 g at birth). Thirty-three families consented, of whom 32 completed all six structured home visits delivered until the infants reached 6 months CA. Parents rated the intervention very positively with a mean satisfaction score of 9.4 (range 0–10), reporting increased confidence, improved understanding of infant signals, and high appreciation for individualized parent reports. Parental outcomes showed increases in parental self-efficacy, reflective functioning (with medium to large effect sizes), and parental sensitivity across both structured interaction tasks. Child outcomes

were similarly encouraging motor development was largely age-appropriate (9% < 5th percentile) and 6% of infants were identified as at risk for socio-emotional difficulties at follow-up.

Overall, the modified six-session TOP program for MP infants was feasible, acceptable, and well-received, and associated with positive parent and child outcomes, providing a strong foundation for broader implementation and future evaluations.

Chapter 4

This randomized pilot study assessed the feasibility and potential effects of the TOP+ intervention, an age-appropriate extension of the Dutch TOP program, targeting VP children at 18–22 months CA. The control group (n = 30) received care as usual. All intervention families (n = 30) received the planned 4–6 home visits, with high parental compliance and satisfaction. All parents evaluated the number of sessions as adequate, the interventionist knowledgeable (95%), and the recommendations of the interventionist's valuable (95%). After baseline adjustment, medium to large effect sizes were found for motor development, cognitive development, and parent–child interaction. Taken together, these findings indicate that the additional TOP+ sessions at toddler age are not only feasible and well-received but are also associated with meaningful developmental gains underscoring the program's potential value for broader implementation and further evaluation.

Chapter 5

This qualitative study explored the information needs of parents of VP infants during the first year at home while enrolled in the Dutch TOP program. Semi-structured interviews were conducted with 10 parents (7 mothers, 3 fathers). Thematic analysis identified three recurring themes across the post-discharge trajectory: 1) Help me understand and cope, reflecting parents' struggles to manage stress and uncertainty; 2) Be fully responsible for my baby, capturing the ambivalence and insecurity felt after discharge when hospital safety nets disappeared and new caregiving challenges emerged; and (3) Teach me to do it myself, highlighting parents' strong desire for tailored, practical, and reassuring guidance to build autonomy and confidence. Parents identified ten priority topics for which they desired more accessible, specific, and developmentally tailored information e.g., motor milestones, feeding transitions, sleep regulation, balancing family well-being. Findings underscore the importance of

blended support: professional, personalized guidance complemented by tailored digital resources that adapt to the infant's developmental stage and parents' evolving needs.

Chapter 6

This systematic app review evaluated the availability, content, quality, and usability of mobile information applications designed for parents of preterm infants after hospital discharge. Apps were included when they provided information for parents on topics and content related to prematurity after hospital discharge. A total of 202 apps were screened. Nine English apps were identified, containing information related to the post-discharge period. This content was not comprehensive for the post-discharge period, mostly covering topics such as vaccinations, follow-up, feeding, sleep, and home oxygen. The study identified significant gaps in the availability and quality of mobile health (mHealth) applications for parents of preterm infants after hospital discharge. Given these substantial gaps in availability, relevance, and usability, the development of a new, co-created app tailored to the specific post-discharge needs of parents of preterm infants proved necessary.

Chapter 7

This mixed-method study developed and evaluated the e-TOP app, a digital information tool designed to address the evolving informational needs of parents of preterm infants during the first year at home. Using a participatory, user-centered design process, the app was co-created with parents, TOP interventionists, and healthcare professionals to ensure clinical accuracy, accessibility, and suitability for users with varying health literacy levels. Content was evidence-based, simplified to B1 language level, and supported by multimedia features. Usability evaluation among 61 parents demonstrated good usability and positive user experiences. However, declining engagement, higher attrition among fathers, and underrepresentation of parents with limited health literacy indicate the need for further developmental refinement to enhance personalization, inclusivity, and long-term engagement.

General discussion

This chapter integrates the key findings from the different studies in this thesis and positions them within the broader context of existing research and the Dutch

health care system. Together, the publications outline a pathway for further innovation, refinement, and sustainable scale-up of early intervention programs.

Improvements in the execution quality of the intervention

A key challenge when interventions scale beyond pilot or trial conditions, is to maintain effectiveness. Interventions that perform well under controlled or pilot conditions often lose effectiveness in real-world settings, where variability in staff training, available resources, and contextual factors such as socio-economic background of parents, can dilute implementation quality and intervention integrity (1-3). Unless deliberate strategies are in place to maintain implementation quality the so-called "scale-up penalty" can occur (4-6). Effective large-scale implementation therefore requires therefore structured processes that combine rigorous fidelity monitoring with flexibility for local adaptation. The Dutch TOP program illustrates this trajectory, moving through carefully staged phases of preparation, piloting, and full implementation, supported by continuous monitoring, and sustainable reimbursement from health care insurers (7). Importantly, follow-up data collected during the scaling-up period (2014–2019), demonstrated that the TOP program continued to produce meaningful developmental benefits for high-risk VP children. They showed significantly better cognitive outcomes and fewer anxious and depressive problems at 2 years' corrected age receiving the TOP program compared with those who did not receive the TOP program (8).

Early in the scaling up process of the TOP program, both the research team and educators were only beginning to recognize the critical role of fidelity measurement in safeguarding program quality. As the program expanded and entered the phase of guided adaptations and further research, the absence of such a tool became a limitation. Especially, considering The Consolidated Standards of Reporting Trials statement (CONSORT) statement, which requires interventions to be reported with sufficient detail to allow replication, including how and when they were administered (9). Extensions such as the Template for intervention description and replication (TIDieR) checklist build on this by further emphasizing the need to describe provider expertise, intervention content, delivery mode, and fidelity strategies (10). Generic fidelity frameworks such as the Framework for Reporting Adaptations and Modifications (FRAME) can support consistency in documenting fidelity and adaptations across different interventions and settings (11). When the main objective is comparative research, cross-intervention evaluation, or theoretical understanding of implementation processes, generic

fidelity measures provide a transferable structure for examining key constructs such as adherence, dose, quality, responsiveness, and differentiation across diverse interventions. For instance, the Pediatric Rehabilitation Observational Measure of Fidelity (PROF), useful in pediatric rehabilitation research, capturing multiple fidelity constructs in a standardized format (12).

However, while such tools provide transferability, their relative insensitivity to intervention-specific key strategies limits their ability to clarify the mechanisms of change. Therefore, an intervention-specific observational fidelity instrument was the best option to monitor delivery quality and detect whether the intended adaptations to the existing TOP program, as described in this thesis, stay consistent with the program's underlying principles (13). Both approaches carry distinct strengths and limitations (Table 8.1). A solution for the future is to create a hybrid fidelity system with 1) common indicators based on generic fidelity constructs such as dosage or adherence allowing comparison across interventions; and 2) intervention-specific items ensuring sensitivity to core components, mechanisms of change, and program integrity.

Table 8.1. Generic versus intervention-specific fidelity measures

Aspects	Generic measures	Intervention-specific measures
Scope	- Broad, across intervention programs	- Tailored to a single program
Focus	- Common domains: adherence, dosage, quality, responsiveness	- Prescribed strategies from theory of change - Competent adherence of interventionists
Strengths	- Cross-program comparability - Captures non-specific processes - Useful for reviews/meta-analysis - Benchmarking, multi-program evaluations	- Monitors actual delivery - Align with program theory - Enhances validity - Useful for training/supervision interventionists
Limitations	- Less sensitive to unique mechanisms - Limited program-level feedback	- Limited comparability across programs - Resource-intensive to develop and monitoring

Scale-up literature consistently shows that programs are most successful when fidelity monitoring is embedded within a wider quality-assurance system that includes ongoing data collection, feedback loops, and structured professional development (14, 15). Building and maintaining such monitoring systems

requires long-term financing, including overhead and infrastructure resources. Without dedicated funding quality assurance, data monitoring systems, and guided adaptations, programs risk implementation drift, reduced effectiveness, and ultimately loss of public or insurance support.

Broadening the target group

Across both studies (the adapted TOP program for MP infants (Chapter 3) and the booster intervention (TOP+) (Chapter 4), parents consistently valued the intervention. Parents of MP infants reported high satisfaction, increased confidence, and strong appreciation for personalized reports, also the families in the TOP+ pilot similarly found the number of sessions appropriate and regarded the interventionists' expertise and recommendations as highly meaningful. Together, these findings suggest that parents do benefit from timely, well-targeted, and responsive support. However, to reduce burden and to safeguard healthcare resources, providing the right support at the right moment.is necessary. An effective preventive approach requires systems that enable early and accurate identification of children and families showing signs of emerging difficulties.

Who benefits most and who needs ongoing intervention

Predictive risk models and reliable screening measures can guide stepped-care decisions. Predictive models are moving beyond neonatal risk factors toward multimodal approaches that integrate clinical, neuroimaging, physiological, and social determinants. Recent evidence illustrates this potential: in a nationwide Swedish cohort of 1062 very preterm infants, Bowe et al. showed that 26 routinely available neonatal and sociodemographic features can identify 93% of children with cognitive delay at 2 years (sensitivity 0.93), with key predictors including gestational age, complications such as IVH, duration of hospitalization, family language and education, prolonged hospitalization, breastmilk feeding (16).

At the same time, recent studies underscore the difficulty of prediction in extremely preterm infants. Van Beek and colleagues reported that although 62% of extremely preterm survivors were unimpaired at 2 years, this proportion dropped to 46% by 5.5 years, with one in three children showing a worse outcome category at school age (17, 18). Longer-term follow-up confirms these risks: by 8 years, up to 25% of children born extremely preterm with both extremely low birth weight and small-for-gestational age status requires special education, most often due to cognitive or behavioral problems (19). Such developmental shifts demonstrate that early assessments, while informative, are only moderately

predictive of later functioning and that trajectories can diverge considerably during early childhood.

While biological and sociodemographic risk factors are important, research consistently shows that the quality of the parenting environment plays a protective role in shaping children's developmental trajectories (20-25). This aligns closely with the vision of the 1001 Critical Days, which emphasizes that the period from conception to age two represents a unique window during which nurturing relationships and emotionally attuned caregiving lay the foundations for lifelong health, learning, and wellbeing (26). The influence of the caregiving context, such as parental sensitivity, responsive interactions is particularly strong for infants at elevated risk, such as those born preterm or with early medical vulnerabilities (27-31).

This underscores the need for additional predictive assessments of parental stress, interaction quality, and family functioning (32). Rather than focusing solely on risk factors, emphasizing protective factors such as parental sensitivity, secure parent–infant interaction, and family resilience provides a more balanced and developmentally informed approach to early identification and intervention for children born preterm.

Adding the e-TOP app to the TOP program

Building on the research–practice gap in accessible and trustworthy post-discharge information for preterm families, we developed a web-based application that strengthen one of the key strategies of the TOP program, namely providing education that empowers parents with knowledge and skills to guide their infant during the first year at home. In line with Bandura's theory (33), such resources enhance parents' beliefs in their ability to positively influence their child's development, which is particularly valuable in the uncertain developmental trajectory of preterm born infants. Parents in our study confirmed this, noting that the app provided reassurance, helped them feel more confident in everyday care, and offered clear, supportive information in an accessible format (Chapter 7).

We used an iterative, user-centered methodology that mirrors the principles of participatory health innovation and technology-supported behavior change frameworks (34-36). Particularly useful was the design thinking principle of iteration and stages, where prototypes are not static endpoints, but evolving solutions shaped by ongoing user feedback (37, 38). Guided by these principles, we combined preparatory steps to understand parental needs (Chapter

5), insights from the systematic app review (Chapter 6), and extensive focus group input from parents and TOP interventionists to define the core requirements of the app. These insights translated into prioritized content themes and refined with experts in neonatology and the Dutch Expertise Centre for reducing health inequities to ensure clarity and accessibility. Working closely with a design company, we then iteratively developed and refined a functional, device-independent web-app (Chapter 7).

The test phase combined quantitative (usage analytics, System Usability Scores (SUS), satisfaction surveys) and qualitative (thinking-aloud sessions, interviews) methods to refine usability and evaluate feasibility. This mixed-method approach is consistent with recommended mixed-method approaches for eHealth development and usability evaluation (39, 40).

In both phases of the study, sampling choices were informed not only by methodological aims but also by pragmatic constraints. The study operated within a fixed timeline of 2 years and a limited budget, which restricted the number of sampling rounds. These constraints reflect the inherent tension described by Palinkas et al. where mixed-method implementation studies must balance methodological ideals with feasibility in real-world service settings (41). Despite these restrictions, the sampling strategy remained appropriate for the exploratory nature of this early-phase digital health development effort.

Future steps: integration and continuity across developmental stages

Future development should aim to include a credible digital information source during the NICU phase and expand beyond the NICU and immediate post-discharge period, to ensure parents can return to the app throughout childhood, not only in the first year of life. Parents' informational needs are not static; they evolve from questions about feeding, sleep, and medical follow-up in infancy to concerns regarding motor skills, cognitive development, schooling, and psychosocial wellbeing in later childhood. To meet these shifting needs, the next step is to create a platform that connects existing applications rather than developing isolated solutions per NICU center or research group.

Building on initiatives such as the Neo Parent app (42) or the Care4neo website (43) in the Netherlands, could help to pool expertise and resources and allow for more efficient content development and support long-term maintenance of the platform. Using the expertise of international research teams such as the developers of the NICU2Home app, which focuses on the transition to home would reduce duplication of efforts and accelerate development (44). However,

health care systems differ substantially, any digital tool must still be adapted to the local context to ensure its relevance, acceptability, and effectiveness.

Broader implications for policy

A challenge for sustaining and scaling parenting interventions, such as the Dutch TOP program, lies in the rapidly changing healthcare landscape and the growing costs of care. In this context, preventive early childhood programs must often justify their added value by demonstrating long-term (cost) effectiveness for the children, their families and society. Economic evaluations of parent–infant support programs suggest that even modest improvements in cognitive and socio-emotional outcomes can offset intervention costs through reduced need for special education, healthcare, and social services later in life (45-48). Recent evidence further strengthens this case: the 2024 Lancet Series on the "next 1000 days" concluded that providing early childhood care and education (ECCE) compared to the societal cost of inaction is estimated at 8–19 times greater than the investment (49, 50). This is likely even higher for children at increased developmental risk, such as those born preterm. Preterm birth is indeed consistently associated with elevated direct healthcare costs in the neonatal period and early childhood, as well as substantial indirect costs related to long-term developmental, educational, and psychosocial support needs (50, 51)

Thus, preventive strategies in preterm populations are also compelling from a cost-effectiveness perspective: they target a high-risk group with elevated baseline costs, where the potential for long-term societal savings is greatest. Yet, beyond the economic argument, the true value of such interventions lies in promoting children's active participation, supporting their developmental potential from the family's perspective, and strengthening parenthood as a foundation for resilience and caregiving (52-56).

Linking follow-up infrastructures to program evaluation

For smaller intervention programs, conducting Return on Investment (ROI) research can be challenging due to resource constraints. Full-scale cost–benefit analyses often require multi-year studies, large samples, and access to diverse administrative datasets, which may be unrealistic for smaller programs. In the Dutch context, one approach to strengthen the economic evaluation and the TOP program impact is to leverage routinely collected follow-up data.

The development and implementation of early interventions for preterm infants require not only scientific rigor but also a culture of learning by doing and sharing. Building effective programs involves sharing insights from both successful and less successful initiatives. This calls for united forces: bringing together research teams, interventionists, and parent organizations such as the Care4 neo. Creating structured spaces for exchange and collective reflection can accelerate progress and strengthen the entire field of early intervention for premature born infants.

Broader implications for research

Feasibility research

The experience of developing and implementing the TOP program has taught us that progress rarely follows a linear path from theory to large-scale randomized controlled trials. Instead, sustainable interventions are shaped over time through cycles of testing, reflection, and refinement. Drawing on this background, we made a conscious decision with the TOP program not to wait for perfect research infrastructures, such as fully validated screening instruments or long-term trial systems, before moving forward. PT infants and families need support, and valuable learning often arises through practice itself.

Within the broader research trajectory toward establishing intervention effectiveness, feasibility research plays a critical and indispensable early role. Before an intervention can be rigorously evaluated in a randomized controlled trial (RCT), its core components must be workable, acceptable, and deliverable within real-world neonatal and early intervention contexts. We found that valuable learning arises during the co-creative design of the interventions. Iterative evaluation allows researchers to identify practical barriers, refine delivery strategies, and ensure the intervention fits the daily realities of home-based support. Our experience underscores that feasibility studies function as a bridge between initial development and full-scale evaluation: they allow researchers to refine content, optimize delivery procedures, identify barriers, and confirm that families and practitioners can engage with the intervention as intended. Only once these elements are functioning and the intervention is shown to be usable, acceptable, and implementable, can an RCT meaningfully assess its true clinical effectiveness. In this way, feasibility work lays the practical and methodological foundation upon which successful RCTs are built, ensuring that the subsequent trial tests a mature, implementable, and contextually grounded intervention rather than an unrefined prototype.

A call for collaborative research

Infants born after 30 weeks of gestation, do not routinely enroll in the structured Dutch national follow-up programs, despite being at increased risk for developmental and learning problems compared to term-born peers. We want to emphasize the importance of structured follow up and early intervention research for this target group. In this context, the Dutch Jeugdgezondheidszorg (JGZ) system (57) can serve as a crucial bridge, as its universal preventive healthcare and developmental monitoring infrastructure provides an opportunity to systematically detect emerging difficulties in MLP children and their families.

The role of digital supports in early intervention services

Digital augmentation provides another pathway for strengthening sustainability and scalability. Purpose-built solutions developed through participatory design and iterative usability testing, such as the e-TOP app, illustrate how digital tools can be successfully embedded in blended delivery models. When combined with professional coaching, digital augmentation not only extends reach under increasing caseloads but also enhances accessibility and equity, ensuring that support is available beyond the confines of scheduled intervention visits. An important aspect is the explicit attention to parents with limited health literacy. This group is often underrepresented in mHealth research underscoring the importance of tailored recruitment approaches, simplified consent procedures, and flexible evaluation methods to ensure that digital health interventions truly reach the often-underserved population.

Implications for education and clinical practice

Traditional professional education in pediatric and rehabilitation disciplines has long been oriented toward direct treatment of the child's impairments. However, early-intervention practice calls for a paradigm shift: from "fixing the child" toward coaching and empowering parents as primary agents of change (58-62). Effective early-intervention programs require well-prepared professionals who act from a clear theory of change, linking their interventions directly to strengthen parental capacities and improve child outcomes. This calls for professional education not limited to theoretical knowledge but also teaches how to apply these principles in real-life settings. In the Dutch context, this direction is supported by the educational program of the EOP. It integrates a strong theoretical curriculum on the consequences of prematurity, early development, and parental stress, with practical training, and supervised home

visits. Drawing from this experience embedding caregiver-coaching modules, video reflections, reflective supervision into pediatric physiotherapy educational programs can enhance this shift.

All studies in this thesis contribute directly to clinical practice, both in education and in implementation. They originated from questions raised in practice, were conducted in close collaboration with practitioners and parents, and brought back into practice to improve daily work. The FITT is integrated into the educational curriculum for pediatric physical therapists, providing explicit rating criteria to guide training and supervision. The e-TOP app acts as an accessible adjunct to the intervention, offering additional support to families and practitioners. The adapted MPT intervention has received a custom billing code regulated by the Dutch Healthcare Authority, which allows insurers and providers to make agreements about healthcare services.

Conclusion remarks

The studies in this thesis, demonstrate that feasibility and pilot work are not only preparatory steps, but essential stages in building interventions that are both meaningful for families and viable for practice. By embedding innovation directly into education and care delivery, the work moves beyond a linear research-to-practice trajectory toward a continuous cycle of co-creation, implementation, evaluation, and adaptation. While effect studies remain crucial to establish long-term impact on infant developmental trajectories and parental outcomes, they are most valuable when conducted on interventions that are carefully co-designed, user-tested, and refined. This ensures that effectiveness is assessed in practice-ready programs that are clearly described, reproducible, and shaped by the real needs and contexts of families.

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Chapter 9

Summary
Nederlandse samenvatting

Summary

Preterm birth profoundly influences children's development, with increased risks for short- and long-term difficulties across health, motor, behavioral, cognitive, and socio-emotional domains. Although neonatal care has advanced considerably, with improved survival and increased implementation of family-centered care in the Neonatal Intensive Care Unit (NICU), challenges often extend beyond the neonatal period and emerge within daily caregiving at home. Many preterm infants continue to show early regulatory vulnerabilities that may affect feeding, sleeping, sustained exploration of their environment, and social-emotional development. For parents, the emotional processing of the preterm birth often begins after discharge. Many parents who felt confident in the NICU describe being insecure at home, when they no longer have immediate access to specialized staff, and when new practical and emotional demands arise in everyday home situations. This combination of increased infant vulnerability and parental stress places parent-infant interaction at risk and underlines the importance of early preventive support for families.

To address this need, the Dutch TOP program is developed as a preventive responsive parenting intervention for parents of very preterm (VP) infants, born before 32 weeks of gestational age. The program is home-based and delivered by pediatric physical therapists during the first year after hospital discharge. The program is grounded in strong evidence demonstrating that responsive parenting interventions can improve motor, cognitive, and socio-emotional child outcomes, strengthen parental sensitivity, and reduce parenting stress. After successful national implementation since 2014, the TOP program now reaches a large proportion of VP infants in the Netherlands (80%) and continues to evolve alongside changing family needs, emerging scientific insights, and new digital possibilities.

This thesis aimed to strengthen the impact of the Dutch TOP program through three approaches: 1) improving execution quality through fidelity monitoring; 2) expanding intervention reach by adapting the program for moderate preterm born (MP) infants and VP born toddlers; and 3) the addition of digital support to strengthen parents' knowledge and understanding.

As early-intervention programs scale beyond pilot and research phases, maintaining stable implementation quality is both essential and challenging. In **Chapter 2** we developed and validated the Fidelity of Implementation Tool for the TOP program (FITT). Three consecutive phases were carried out resulting

in a tool consisting of adherence and competence subscales plus a general impression score. Results showed strong interrater reliability and good internal consistency. In addition, the tool demonstrated good discriminative ability, effectively capturing meaningful variation in provider performance.

The original TOP program focuses on the highest-risk VP population. However, MP infants, born between 32–35 weeks gestation, represent a much larger group who also display elevated risks for developmental impairment compared to term-born peers. **Chapter 3** evaluated the feasibility and acceptability of an adapted six-session TOP program for MP infants. The study included 33 families, of whom 32 completed the intervention. Parents reported high satisfaction, increased confidence, and greater understanding of their infant's signals. Parent and child outcomes were encouraging. These findings provide a strong foundation for broader implementation and future evaluations.

Despite early interventions in infancy, many VP children continue to face emerging difficulties in toddlerhood. As children transition into new stages, they are expected to acquire more complex cognitive, language, motor, and socio-emotional skills, which can reveal challenges that were not yet apparent in early infancy. This highlights the importance of ongoing monitoring and timely booster interventions that align with each new developmental phase. **Chapter 4** examined the feasibility and potential effects of TOP+, a structured booster program for VP children delivered at 18–22 months corrected age. In this randomized pilot trial, families in the intervention group received 4–6 home visits focusing on motor skills, parent-child interaction, and regulatory functioning. Parents rated the content and number of visits as appropriate, valued the individualized parent report with strength-based recommendations and showed high engagement. Preliminary effects at 24 months showed promising improvements in child motor and cognitive development, reductions in behavior problems, and improved parent-child interaction quality.

Chapter 5 addressed a critical gap in existing resources by exploring the evolving informational needs of parents of VP infants during the first year at home. Semi-structured interviews with 10 parents identified three dominant themes: 1) Help me understand and cope, reflecting the emotional and cognitive challenges following NICU discharge; 2) Help me to be fully responsible for my baby, capturing parents' struggle with stress, insecurity, and role transition; and 3) Teach me to do it myself, highlighting their desire for practical, developmentally attuned guidance supporting autonomy and confidence. In addition, parents identified ten priority topics for which they desired more accessible,

specific, and developmentally tailored information, e.g., motor milestones, feeding transitions, sleep regulation, balancing family well-being. The findings provided input for formulating the requirements of a mobile health application.

In addition to the interviews and established design requirements (Chapter 5), existing digital resources were explored to prevent overlap with existing mobile health initiatives. **Chapter 6** describes the systematic review of mobile applications for parents of preterm infants after discharge. Screening 202 apps yielded only nine with post-discharge content, none offering comprehensive, developmentally tailored information for the full first year at home. The overall quality of the apps was only acceptable. Although the reliability of the information was close to good, the understandability of the apps was moderate. This review identified significant gaps in the availability and quality of mobile health applications for parents of preterm infants after hospital discharge.

Chapter 7 describes the development and evaluation of the e-TOP app, designed to deliver trustworthy, and specific information to parents of VP infants. Using participatory design, the development process included parents (including those with limited health literacy), TOP interventionists, pediatric physical therapists, and content experts. The app contained easy-to-understand text, illustrative photos, animations, and videos. Usability testing in 61 parents demonstrated high usability and positive user experiences. However, analytics showed declining engagement over the period of 6 months, limited uptake among fathers, and underrepresentation of parents with low health literacy, a persistent equity concern. These findings highlight the need for enhanced personalization, and integration of the app in the TOP intervention pathway.

Conclusion

A central insight from this thesis is that the development and innovation of early-intervention programs do not follow a linear or predetermined path. Rather, each deliverable and result, including the Fidelity tool, adapted interventions for MP infants and toddlers and the e-TOP application, was shaped through iterative cycles of co-creation, testing, adaptation, and refinement. Crucially, these processes were informed by the continuous participation of relevant stakeholders, such parents, health care professionals, and researchers, whose insights shaped both the design of new components and the modification of existing interventions.

The early pilot tests generated actionable insights such as identification of practical barriers, usability challenges, and mismatches with parental needs.

In addition, they provide promising indications of positive effects on child and parent outcomes. This practice-oriented and feasibility-focused research approach increases the likelihood of successful implementation in practice and ensures that future effectiveness studies are meaningful for preterm children and their parents.

Nederlandse samenvatting

Vroeggeboorte heeft grote gevolgen voor de ontwikkeling van kinderen en gaat gepaard met een verhoogd risico op problemen op het gebied van gezondheid, motoriek, gedrag, cognitie en sociaal-emotioneel functioneren. Hoewel de neonatale zorg sterk verbeterd is, met betere overlevingskansen en meer aandacht voor gezinsgerichte zorg tijdens de ziekenhuisopname, ontstaan er vaak nieuwe uitdagingen zodra gezinnen thuis zijn. Te vroeg geboren kinderen hebben in de eerste periode na ontslag vaak regulatieproblemen. Deze kunnen zich uiten in problemen met bijvoorbeeld slapen, drinken, interactie met de omgeving en de sociaal-emotionele ontwikkeling. Voor ouders begint de emotionele verwerking van de vroeggeboorte vaak pas na ontslag uit het ziekenhuis. Veel ouders die zich op de Neontale Intensive Care afdeling (NICU) zelfverzekerd voelden, geven aan zich thuis onzeker te voelen. Dit hangt samen met het wegvallen van directe toegang tot zorgprofessionals en nieuwe praktische en emotionele uitdagingen in het dagelijks leven met hun baby. De combinatie van verhoogde risico's op ontwikkelingsproblemen en toegenomen ouderlijke stress zet de ouder-kindinteractie onder druk en benadrukt de noodzaak van vroege ondersteuning.

Tegen deze achtergrond werd het Nederlandse TOP-programma ontwikkeld als een preventief interventieprogramma voor zeer prematuur geboren kinderen (VP; zwangerschapsduur < 32 weken) en hun ouders. Het programma heeft tot doel de vroege ontwikkeling van het kind te bevorderen door het versterken van responsieve ouder-kindinteractie en bevorderen van de zelfregulatie en het faciliteren van de ontwikkeling van het kind. De interventie wordt gedurende het eerste jaar na ontslag thuis bij het gezin uitgevoerd door gespecialiseerde kinderfysiotherapeuten. Het TOP-programma is onderbouwd door wetenschappelijke evidentie waaruit blijkt dat interventies gericht op responsief ouderschap de motorische, cognitieve en sociaal-emotionele ontwikkeling van kinderen kunnen verbeteren, ouderlijke sensitiviteit kunnen versterken en stress bij ouders kunnen reduceren. Sinds de landelijke implementatie in 2014 bereikt het TOP-programma ongeveer 80% van alle VP-kinderen in Nederland. Het programma blijft zich ontwikkelen op basis van veranderende gezinsbehoeften, nieuwe wetenschappelijke kennis en digitale mogelijkheden.

Het doel van dit proefschrift was de impact van het TOP-programma te vergroten door 1) het verbeteren van de kwaliteit van de uitvoeringspraktijk; 2) het aanpassen van het TOP-programma voor matig vroeggeboren kinderen (MP)

en het ontwikkelen van een boosterinterventie voor VP-dreumesen; en 3) het toevoegen van digitale ondersteuning ter versterking van kennis en inzichten van ouders.

Bij het opschalen van vroege interventieprogramma's na de pilot- en onderzoeksfase is het belangrijk, maar ook complex om de kwaliteit van de uitvoering (fidelity) te borgen. **Hoofdstuk 2** beschrijft de ontwikkeling en validatie van de Fidelity of Implementation Tool for the TOP program (FITT). Drie opeenvolgende fasen leidden tot de ontwikkeling van een instrument met twee subschalen: werken volgens de kernstrategieën en het competent uitvoeren van de interventie, aangevuld met een score voor de algemene indruk. De resultaten lieten een sterke inter-beoordelaarsbetrouwbaarheid en een goede interne consistentie zien. Het instrument kon relevante verschillen in uitvoering tussen professionals identificeren.

Het oorspronkelijke TOP-programma richt zich op de hoog risicogroep, VP-kinderen. MP-kinderen, geboren na 32–35 weken zwangerschap, vormen een aanzienlijk grotere groep met eveneens verhoogde risico's op ontwikkelingsproblemen vergeleken met à term geboren leeftijdsgenoten. In **Hoofdstuk 3** wordt de haalbaarheid en aanvaardbaarheid geëvalueerd van een aangepaste versie van het TOP-programma voor MP-kinderen, bestaande uit zes sessies. In totaal namen 33 gezinnen deel waarvan 32 het programma afrondden. Ouders gaven aan dat zij zeer tevreden waren met het programma, meer zelfvertrouwen hadden en de signalen van hun kind beter begrepen. Ouder- en kinduitkomsten waren positief. Deze bevindingen vormen een sterke basis voor bredere implementatie en toekomstige evaluaties van dit programma.

Ondanks vroege interventies in de babytijd, krijgen veel VP-kinderen in de peuterleeftijd te maken met nieuwe problemen. Naarmate kinderen nieuwe ontwikkelingsfasen bereiken, zijn complexere cognitieve, taal-, motorische en sociaal-emotionele vaardigheden nodig, waardoor problemen zichtbaar kunnen worden die eerder nog niet opvielen. Dit benadrukt het belang van voortgezette monitoring en tijdige boosterinterventies die aansluiten bij elke nieuwe ontwikkelingsfase.

Hoofdstuk 4 beschrijft de haalbaarheid en potentiële effecten van TOP+, een gestructureerd boosterprogramma voor VP-kinderen aangeboden op de gecorrigeerde leeftijd van 18–22 maanden. In deze gerandomiseerde pilotstudie kregen gezinnen in de interventiegroep 4–6 huisbezoeken gericht op motoriek, ouder-kindinteractie en zelfregulatie. Ouders waarderen de inhoud en het aantal bezoeken, en waren sterk betrokken. De resultaten op 24 maanden lieten

veelbelovende verbeteringen zien in motorische en cognitieve ontwikkeling, gedragsproblemen en ouder-kindinteractie.

Hoofdstuk 5 richt zich op het verkennen van de veranderende informatiebehoeften van ouders van VP-kinderen in het eerste jaar thuis. Op basis van semigestructureerde interviews met tien ouders kwamen drie thema's naar voren: 1) help me begrijpen en ermee om te gaan, gericht op de emotionele en cognitieve uitdagingen na NICU-ontslag; 2) help me volledig verantwoordelijk te zijn voor mijn baby, waarin ouders hun worsteling met stress, onzekerheid en roltransitie beschreven; en 3) leer me het zelf te doen, waarin ouders hun behoefte aan praktische, ontwikkelingsgerichte ondersteuning benadrukten. Daarnaast benoemden ouders tien onderwerpen waar ze informatie over zouden willen, zoals motorische mijlpalen, voeding, slapen en balans in het gezinsleven. Deze resultaten vormden input voor de ontwerpvereisten van een mobiele eHealth-applicatie.

Als aanvulling op de interviews en de vastgestelde ontwerpeisen in Hoofdstuk 5, is onderzocht welke digitale middelen al beschikbaar zijn om overlap met bestaande initiatieven te voorkomen. **Hoofdstuk 6** bevat een systematische review van mobiele apps voor ouders van prematuur geboren kinderen. Van de 202 gescreende apps bleken slechts negen apps informatie te bevatten over de periode na thuiskomst uit het ziekenhuis. Geen enkele app bevatte specifieke informatie voor het gehele eerste levensjaar. De algehele kwaliteit van de apps was acceptabel. Hoewel de betrouwbaarheid van informatie goed was, was de begrijpelijkheid matig. De review brengt duidelijke tekortkomingen aan het licht in de beschikbaarheid en kwaliteit van mHealth-toepassingen voor ouders na ontslag uit het ziekenhuis.

Hoofdstuk 7 beschrijft de ontwikkeling en evaluatie van de e-TOP-app, bedoeld om betrouwbare en gerichte informatie te bieden aan ouders van VP-kinderen. Bij het ontwikkelproces waren ouders (ook met beperkte gezondheidsvaardigheden), (TOP) kinderfysiotherapeuten, en inhoudelijke experts betrokken. De app bevatte begrijpelijke tekst, illustratieve foto's, animaties en video's. Tests met 61 ouders lieten zien dat de app gebruiksvriendelijk was en als zeer positief werd ervaren. Analyse van het gebruik toonde een dalende gebruikersactiviteit tijdens de periode van 6 maanden en beperkte deelname aan het onderzoeksdeel van de studie door vaders en ouders met beperkte gezondheidsvaardigheden. Deze bevindingen benadrukken het belang van verdere personalisatie en de integratie van de app in het zorgtraject.

Conclusie

Een centrale conclusie van dit proefschrift is dat het ontwikkelen en innoveren van interventieprogramma's geen lineair of vooraf bepaald traject volgt. Alle resultaten van deze studies, het fidelity-instrument, de aangepaste interventies voor MP-kinderen en VP-dreumesen en de e-TOP-app, zijn stap voor stap ontwikkeld via iteratieve cycli van co-creatie, toetsing en bijstelling. Het structureel betrekken van stakeholders, waaronder ouders, zorgprofessionals en onderzoekers, was hierbij van groot belang. Hun inzichten hebben geholpen bij het ontwerp van nieuwe interventieonderdelen en het aanpassen van bestaande interventies.

De pilotstudies bieden inzichten in praktische knelpunten, de gebruiksvriendelijkheid van de interventies en de mate waarin ze aansluiten bij de behoeften van de ouders. Ze leveren daarnaast veelbelovende aanwijzingen op voor positieve effecten op kind- en ouderuitkomsten.

Deze wijze van praktijkgericht en op haalbaarheid gericht onderzoek, vergroot de kans op succesvolle implementatie in de praktijk en zorgt ervoor dat toekomstige effectstudies betekenisvol zijn voor vroeggeboren kinderen en hun ouders.

Chapter 10

Dankwoord
List of abbreviations
List of contributing authors and affiliations
List of author contributions
PhD portfolio
Curriculum vitae

Dankwoord

Ouders die te maken krijgen met een vroeggeboorte beginnen aan een weg die anders verloopt dan vooraf gedacht. Onderweg is er onzekerheid en kwetsbaarheid, het voortdurend aanpassen aan nieuwe omstandigheden staat centraal. Promoveren vraagt net als ouderschap na een vroeggeboorte om het loslaten van verwachtingen, genieten van het moment, leren vertrouwen op jezelf en vertrouwen op steun van anderen.

Deze parallel heeft mij geholpen om het onderzoekswerk niet los te zien van het leven zelf. Het heeft mijn begrip verdiept van de betekenis van nabijheid. Het heeft me geleerd steun te kunnen ontvangen. En daarmee te kunnen groeien als moeder van Hannah en Fleur, als vriendin, onderzoeker, TOP-kinderfysiotherapeut en als mens.

It took a village to complete this thesis.

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Al deze mooie studies zijn gedaan in samenwerking met ouders en zeer betrokken **TOP-kinderfysiotherapeuten**. Speciaal wil ik de **ouders** bedanken die ik in de afgelopen jaren heb mogen begeleiden. Hun kennis en inzichten hebben mij geholpen bij dit proefschrift.

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weet ik dan wat je ervan vindt. En je levensmotto; Het komt goed. Soms zo irritant maar oh zo waar.

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List of abbreviations

ADS	The Massie-Campbell Attachment during Stress Scale
AIMS	Alberta Infant Motor Scale
ASQ-3	Ages and Stages Questionnaire, 3rd edition
ASQ-SE2-NL	Ages and Stage Questionnaire Socio-Emotional Dutch version, 2nd edition
BSID-III-NL	Bayley Scales of Infant and toddler Development, Third Edition, Dutch Version
BPD	Bronchopulmonary dysplasia
BW	Birth weight
CA	Corrected age
D/C	Discharge
DT-P	Distress Thermometer for Parents
EAS	Emotional Availability Scales
EOP	Expertise center for developmental support for premature infants
FCC	Family-centered care
FITT	Fidelity of Implementation Tool TOP program
FT	Full term
GA	Gestational age
IBAIP	Infant behavioral assessment and intervention program
IC	Informed consent
ITSEA	Infant toddler social and emotional assessment
IVH	Intraventricular hemorrhage
IQR	Interquartile range
LBQ	Lexi-list perceptive language quotient
LHL	Limited health literacy
LOS	Length of hospital stay
MARS	Mobile App Rating Scale
NEC	Necrotizing enterocolitis
NICU	Neonatal Intensive Care Unit
M	Mean
mHealth	Mobile Health
MP	Moderate preterm
MLP	Moderate-and late preterm
N/A	Not applicable

NEC	Necrotizing enterocolitis
PEMAT-AV	Patient Education Materials Assessment Tool for Audiovisual Materials
PPT	Pediatric physical therapist
PRF/PRFQ	Parental Reflective Functioning Questionnaire
PVL	Periventricular leukomalacia
PRISMA	Preferred Reporting Items for Systematic reviews and Meta-Analyses
PSE	Parental self-efficacy
PT	Preterm
RCT	Randomized control trial
SD	Standard deviation
SES	Socioeconomic status
SENR	Maternal Self-Efficacy in the Nurturing Role
SGA	Small for gestational age
STIPP	Stedelijk Transmuraal Interventie Programma voor Prematuur geboren kinderen en hun ouders
SUS	System usability scale
TOC	Theory of change
TOP	Transmural developmental support for preterm children and their parents
VP	Very preterm
VPT	Very preterm
PT	Preterm

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Data collection: MF, EV
Data interpretation: MF, EV, MJ
Drafting first version of the manuscript: MF
Critical revision and editing of the manuscript: all researchers involved.

Chapter 3: Feasibility of a Dutch post-discharge parenting intervention (TOP program) for moderate preterm born infants

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Data collection: MF, EM
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Chapter 4: Feasibility of a preventive parenting intervention for very preterm children at 18 months corrected age: A randomized pilot trial

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Data collection: MF, KK, DM
Data interpretation: MF, KK, DM, AL, PS, MJ
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Chapter 5: Parents' information needs during the first year at home with their very premature child; a qualitative study

Researchers involved: M. Flierman (MF), D. Bossen (DB), R.A. de Boer (RB), E. Vriend (EV), F. van Nes (FN), A.H. van Kaam (AK), R.H.H. Engelbert (RE), M. Jeukens-Visser (MJ)
Study concept and design MF, DB, RB, EV, MJ, FN, RE
Data collection: MF, RB
Data interpretation: MF, DB, RB, FN, EV, MJ
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Chapter 6: Evaluating mobile information applications for parents of preterm infants after hospital discharge: A systematic app review

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Data collection: MF, MJ, RG
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Critical revision and editing of the manuscript: all researchers involved.

Chapter 7. An information app (e-TOP) to support parents' information needs during the first year at home after preterm birth: Development and usability study

Researchers involved: M. Jeukens-Visser (MJ), M. Flierman (MF), E.L. Moller (EM), V.L. Vijn (VV), R.H.H. Engelbert (RE), A.H. van Kaam (AK), D. Bossen (DB)
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Data interpretation: MF, VV, DB, EM, MJ, RH
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PhD portfolio

Name PhD student	Monique Flierman
PhD period	2021-2025
Names of PhD supervisors	Prof. dr. R.H.H Engelbert Prof. dr. A.H.L.C. van Kaam
Names of PhD co-supervisors	Dr. D. Bossen Dr. M. Jeukens-Visser

PhD training

Year		ECTS
	General courses	
2022	Basiscursus regelgeving en organisatie voor klinische onderzoekers (e-BROK)	1.5
2023	Qualitative research	1.9
	Educational programs, Masterclasses, webinars, workshops	
2021-2023	RINO, Utrecht, the Netherlands *Infant Mental Health opleiding* Module 1: Visie en vaardigheden gericht op ouder-kindrelatie in context Module 2: Triage en diagnostiek in de ouder-kind relatie Module 4: Kortdurende focusgerichte interventies, gericht op (aankomend) ouderschap en/of ouder Module 7: IMH: Integreren van IMH in de werksituatie	 2.0 2.0 2.0 1.0
2021-2023	Organizing and supervising focus group meetings	2.0
2021	Workshop EOP-HvA, Amsterdam, the Netherlands *Teaching intervention and research protocol*	1.0
2023	Webinar Building resilience through play. Center of the Developing Child, Boston, USA	0.5
2024	Valorisatiebijeenkomst Pebbles, HvA, Utrecht, the Netherlands	0.5
	Oral presentations	
2021	Werkgroep regionale neonatologie, SCEM, Bergen, the Netherlands *Zoektocht van ouders en zorgprofessionals naar de juiste hulp voor ouder en kind*	0.5
2022	Jaarcongres NVFK/NVK, Arnhem, the Netherlands *TOP-programma; implementatie, borging en innovatie*	0.5
2022	Dutch working group of neonatal follow up (LNF), Veldhoven, the Netherlands *Digitale informatie ondersteuning voor ouders met een vroeggeboren kind*	0.5
2022	Hogeschool van Amsterdam, the Netherlands *Study pitch: Development of the e-TOP app*	0.3

Year		ECTS
2023	Amsterdam Kinder symposium, Amsterdam, the Netherlands *The development of the e-TOP information app for parents of very and moderate preterm-born infants*	0.5
2023	World Congress Infant Mental Health (WHAIM), Dublin, Ireland *There is an app for that*	0.5
2024	Dutch working group of neonatal follow up (LNF), Nijmegen, the Netherlands *Development and evaluation of the e-TOP app Feasibility TOP program for moderate preterm born infants*	0.5
2024	EOP symposium, Zeist, the Netherlands *De reis van het EOP en het TOP-programma*	1.0
2024	Nidcap Federation International, Toulouse, France *Early Developmental Care; TOP program*	0.5
2025	Werkgroep regionale neonatologie, SCEM, Bergen, the Netherlands *TOP-programma voor matig en laat geboren prematuren*	0.5
2025	Conference of Society on Early Intervention (ISEI), Lissabon, Portugal Oral presentation: *Tools to Support Communication about Parental Beliefs and Information Needs in Early Intervention*	1.0
	Other research-related accomplishments	
2021	Co-authorship of the SIA-RAAK MKB grant proposal	1.5
2022	Co-authorship of the Stichting Achmea Gezondheidszorg (SAG) funding application	1.0
2023	Co-authorship Projectidee ZonMw Doelmatigheid van een responsief ouderschapsprogramma voor matig prematuur geboren kinderen	1.0
2024	Authorship funding applications NZA-Facultatieve Prestatie Code	2.0
2023	Revising EFCNI standard on Post-discharge responsive parenting programs	2.0
	Publications magazines	
2024	IMPACT Magazine	0.5
2025	VROEG	1.0
2025	Kleine maatjes	0.5
	Attendance (inter)national conferences	
2022	Werkgroep regionale neonatologie, SCEM, Bergen, the Netherlands	0.5
2022	Jaarcongres NVFK/NVK, Arnhem, the Netherlands	0.5
2022	Dutch working group of neonatal follow up (LNF), Veldhoven, the Netherlands	0.5
2022	Amsterdam Kinder symposium, Amsterdam, the Netherlands	0.5
2024	World Congress Infant Mental Health (WHAIM), Dublin, Ireland	1.5
2024	Dutch working group of neonatal follow up (LNF), Nijmegen, the Netherlands	0.5
2024	EOP De reis van de prematuur, Zeist, the Netherlands	0.5
2025	Meeting NIDCAP Federation International, Toulouse, France	1.0
2025	Werkgroep regionale neonatologie, SCEM, Bergen, the Netherlands	0.5
2025	Conference of Society on Early Intervention (ISEI), Lissabon, Portugal	1.5

Year		ECTS
	Teaching	
2020	NIDCAP Masterclass, ErasmusMC, Rotterdam *Ontwikkelingsondersteuning rondom de uitgerekende datum*	0.5
2021–2024	TOP opleiding, teaching and supervising, EOP Amsterdam	2.0
2021	Supervising fellow researcher	1.0
2023	Supervising student Pedagogische en Onderwijswetenschappen Masterthesis	1.5
2023	Supervising students (2) Exercise therapy, Bachelor year 3	1.5

Curriculum vitae

Monique Flierman was born on March 1, 1968, in Ameide, the Netherlands. After completing her secondary education (atheneum) at De Bruijne Lyceum in Utrecht in 1987, she pursued a degree in Physical Therapy at Thim van der Laan School of Physiotherapy in Utrecht, graduating in 1991. From the beginning of her professional career, she developed a strong interest in children's development and the influence of environmental and relational contexts on developmental trajectories. Alongside her clinical work as a physical therapist, this interest led her to start a master's program in Educational Sciences at Utrecht University and she graduated in 1994.

To broaden her clinical experience, Monique worked for four years at Riley Children's Hospital in Indianapolis, USA. This period significantly deepened her understanding of the impact of illness on children and their families and further strengthened her commitment to family-oriented and developmentally supportive care.

After returning to the Netherlands, she worked in primary care pediatric physiotherapy practice in Bleiswijk and at Erasmus MC–Sophia Children's Hospital in Rotterdam, where she combined clinical work with a specialization in Pediatric Physical Therapy. She obtained her postgraduate diploma at the Hogeschool van Utrecht in 2001. During this time, her interest increasingly focused on neonatology specifically in family-centered care approaches. She was actively involved for seven years in development and implementation of the Newborn Individualized Developmental Care and Assessment Program (NIDCAP) in the Netherlands.

Her broad interest in the contextual factors affecting vulnerable children and families, continued at Flexus Jeugdplein in Rotterdam. where she engaged in the development and implementation of a comprehensive, multi-year program focused on effectiveness research and methodological innovation within youth services in Rotterdam.

In 2014, she started working at the AmsterdamUMC for the Expertise Center for Developmental support for premature born infants (EOP). Her role focused

on three core responsibilities: developing educational programs, ensuring quality assurance for the TOP program, and contributing to policy development.

In subsequent years, Monique was given the opportunity to combine her professional interest in practice-based research with a PhD position. She began her doctoral journey in 2021 at the Amsterdam Reproduction & Development Research Institute. During her PhD trajectory, she combined her research activities with clinical work as a TOP-pediatric physical therapist and executive leadership as director of the EOP.