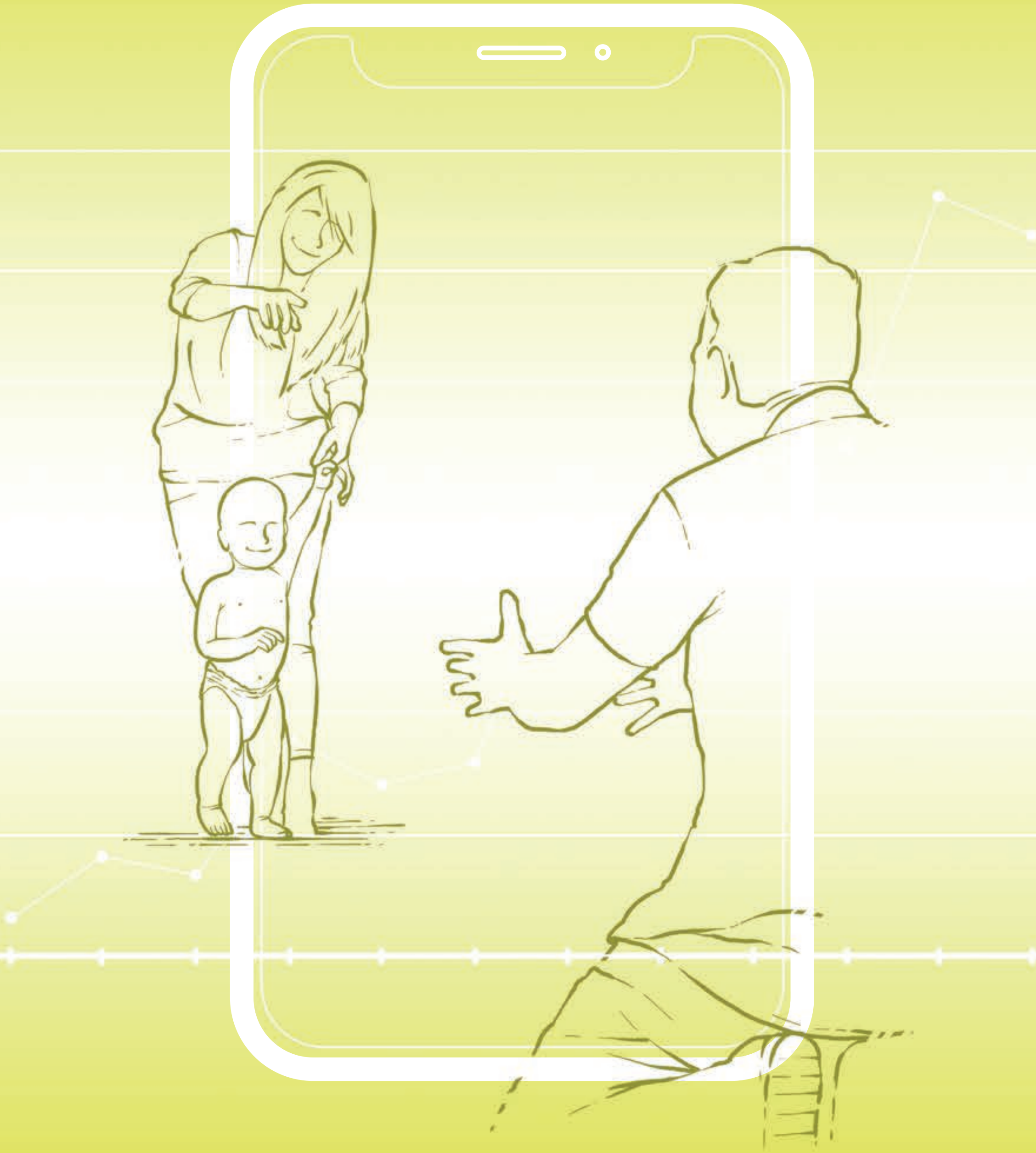
From birth to independent walking

Gross motor trajectories of infants captured by home videos made by parents



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From birth to independent walking: Gross motor trajectories of infants captured by home videos made by parents

Vanaf geboorte tot het zelfstandig lopen: Grof-motorische ontwikkelingstrajecten van jonge kinderen in beeld gebracht met homevideo's gemaakt door ouders

(met een samenvatting in het Nederlands)

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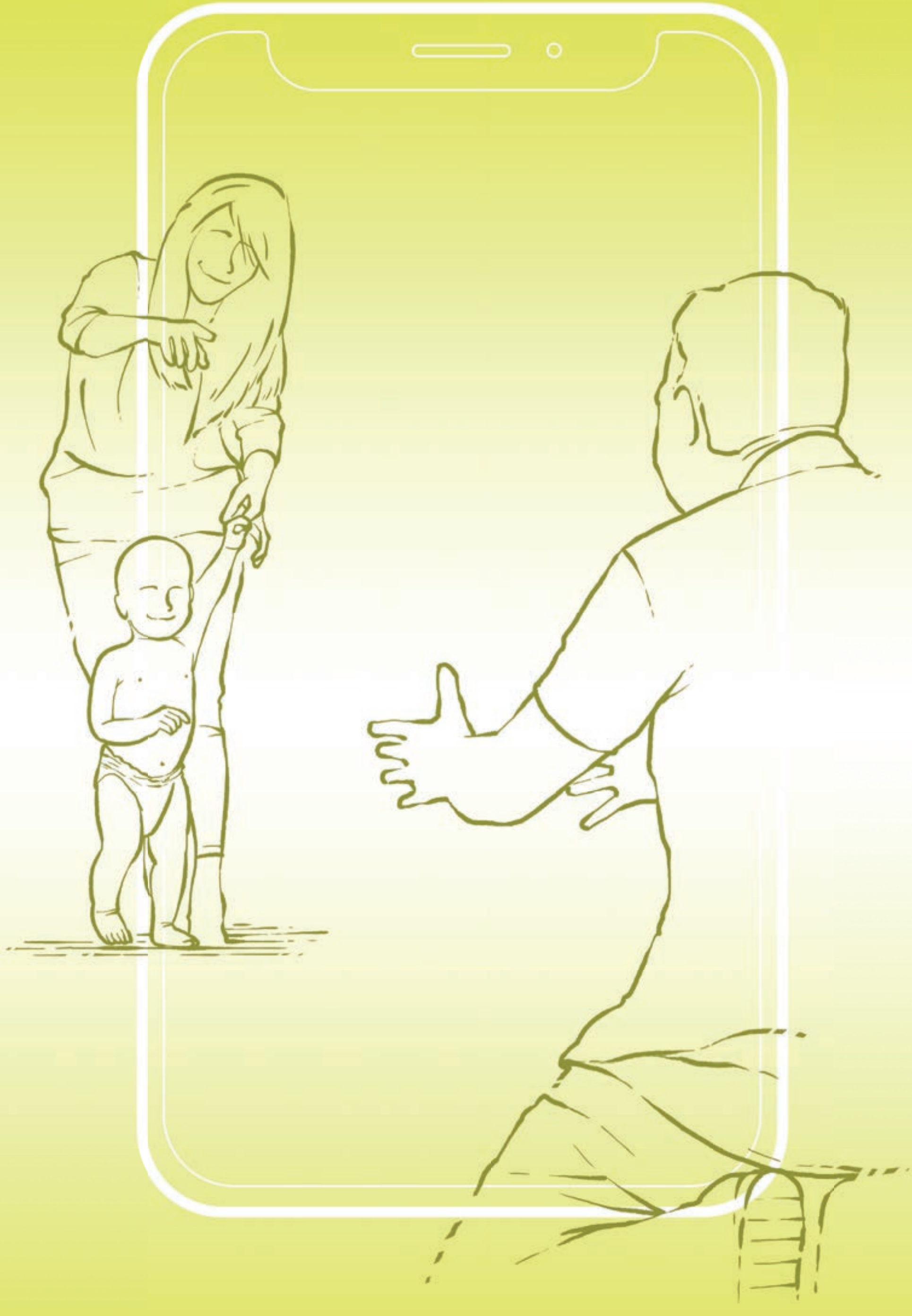

Chapter 1

General introduction

Gross motor development

Infancy is a time of huge leaps in motor development. In the first two years of life, parents closely follow the acquisition of new skills in their baby's motor repertoire. For health care professionals, gross motor development is an important indicator of health and general development.[1] It provides an early insight into the integrity of the brain.[2] Not only does observing gross motor development in infancy inform about the baby's instantaneous state: several studies have shown the predictive value of early motor outcomes on later functioning in other developmental domains, such as cognition,[3-5] and language development.[6,7]

A child's motor development is defined as the change in motor behaviour experienced over the span of childhood life.[8] Gross motor development refers to movements that require the use of large muscle groups that coordinate body movements to perform activities such as: maintaining the head in the midline, rolling, creeping, crawling, sitting, and independent walking.[9]

Early theories of motor development emphasized the idea of uniformity and linearity in the rate and sequence of the achievement of new motor milestones. These theories were based on the premise that motor development was mainly driven by the maturation of the central nervous system.[10] From this idea, many normative descriptions and charts reflecting the typical ages of the achievement of milestones have emerged that are still in use today.

However, in recent decades, numerous studies have shown great variability in the age and sequence of achieving gross motor milestones among infants.[11-13] Variability has also been observed with individual infants repeatedly executing the same movement with small differences in each repetition. These findings have given way to a new paradigm. The dynamic systems theory, as applied to motor development, relates the variability in gross motor development to ongoing interactions between the child (behavioural and physical aspects), the environment, and the motor task.[14] This theory is characterized by the idea of a self-organizing system in which all subsystems are equally important. A small change in one subsystem can trigger a cascade of changes in other subsystems and may result in new motor behaviour. This assumption fits the idea that the development of new motor behaviour is a non-linear process defined by intra- and inter-individual variability.[11,15]

Another theory that underlines the role of factors related to the emergence of new motor behaviour is Newell's theory of constraints (Figure 1.1).[16] He suggested that motor development is shaped by the interaction of individual, environmental, and task-based constraints.[17] The overall movement pattern changes if the constraints are changed, these being features of the individual, of the environment, or of the task that either limit, contain or help shape the development. In the motor behaviour of infants, the surface on which the infant is moving can be seen as an environmental constraint. For example, the infant might slide forward on the belly on a smooth floor but change the method of locomotion to crawling when on a rough carpet.

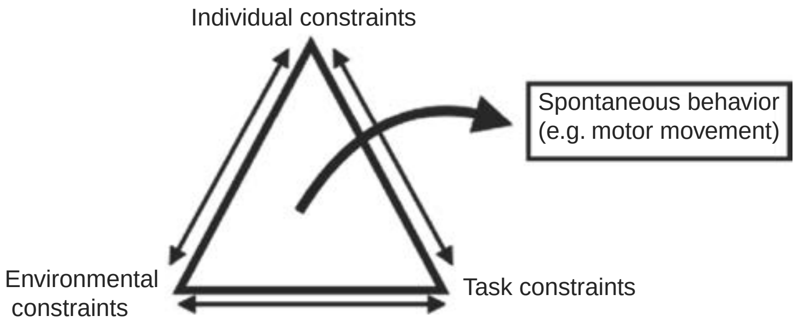

Figure 1.1 Newell's theory of dynamic systems.
Reprinted with permission of Colombo-Dougovito, A. M. (2016). The role of dynamic systems theory in motor development research: how does theory inform practice and what are the potential implications for autism spectrum disorder?. International Journal on Disability and Human Development, 16(2), 141-155.

It thus emerges that gross motor development is the outcome of the interaction of the growing and maturing infant, the motor tasks, and the environment, a very complex and dynamic process in which many factors interact, and whose outcome is difficult to predict. Understanding motor development is important in two ways:

1. Variability between and within infants defines the developmental process but also challenges the early detection of delays in gross motor development. As a consequence, early intervention might not always be provided for those children who would benefit most from it, while on the other hand, over-referral and unnecessary concerns are common.[18]

2. A better understanding of the variability in gross motor development can support researchers and clinicians in understanding how the self-organizing system 'learns' new motor behaviour. Insight into the impact that factors have on this dynamic process provides a starting point for effective interventions that aim to improve infant motor development.

The main aim of the studies presented in this thesis is to gain insight into the inter-individual variability that is present in the gross motor development of typically developing (TD) infants. This is to be achieved by 1) introducing a home-video method to facilitate longitudinal data collection to model a gross motor growth curve, and 2) by studying factors that are associated with gross motor development from birth to independent walking.

The assessment of gross motor development

In the Netherlands, with about 165,000 new-borns born each year,[19] a procedure of national developmental monitoring aims to identify as early as possible infants at risk of delayed (motor) development. Early detection is important because of the plasticity of the young brain. A large body of evidence supports the idea that the first 1,000 days of life provide an important window of opportunity in which early interventions can exert the most impact to optimize development.[20] Thus, from birth onwards, physical growth and development are regularly monitored in well-baby clinics. In the case that signs indicate a delayed or deviant motor development, parents are referred to a Paediatric Physical Therapist (PPT) for diagnostics and early intervention. The main goal of the PPT in early intervention is to improve the development and functioning of the child.[21]

The Alberta Infant Motor Scale

To assess gross motor development in the first two years of life, several measurement tools are available. The studies presented in this thesis revolve around the Alberta Infant Motor Scale (AIMS).[22] This measurement tool was developed to assess the gross motor maturation of TD infants from 0 to 19 months by observing spontaneous motor behaviour. The psychometric properties of the AIMS are considered to be excellent.[23] The scale consists of 58 items divided into four subscales of positions in which the infant is observed: prone, supine, sitting, and standing. The items are arranged in the order of development that is most common among infants. A raw score can be calculated by adding up the scores on the observed items and the preceding items that are considered to have been mastered. The raw score can be converted into a percentile score and a z-score, using reference values available from 2,202 Canadian infants.[22] In 2014, a re-evaluation was carried out which indicated that these reference values were still adequate for infants living in Canada.[25] In the absence of Dutch norms, these Canadian norms are also applied by PPTs in the Netherlands. In past years, the cross-cultural validity of these reference values has attracted interest because evidence emerged that Dutch infants progress at a slower developmental pace than do Northern American infants.[26,27] Very recently, Dutch AIMS norms were established with 1697 infants confirming this delay.[28]

Challenges in the assessment of gross motor development

Monitoring infants over time

This thesis started with the question of whether videos made by parents at home could later be used by PPTs to assess infant motor development. The use of home videos could lessen the (travel) burden that multiple assessments in follow-up clinics and in longitudinal research place on infants and parents. Particularly for younger parents, it is nowadays quite customary to take smartphone photographs and videos of their offspring. A UK study in 2017 reported that the average British parent shares almost 1,500 images of their child online before the fifth birthday.[29] Furthermore, virtually all parents in the Netherlands have access to a smartphone (the mean percentage of Dutch persons (18–45 years) with access to a smartphone was reported to be 98.8%.[30]

The use of home videos in the context of motor development is not new. Several retrospective studies have used home videos to learn about the early signs of autism in the (motor) behaviour of infants from 0 to 24 months.[31-33] The opportunities of home videos made by parents were also studied with the General Movements Assessment (GMA).[34,35] Recent contextual events, e.g., the COVID-19 pandemic, underline the need to monitor infants without physical contact and seem to be driving and accelerating these innovations.

In 2013, the GODIVA research project (**G**ross m**O**tor **D**evelopment of **I**nfants using home **V**ideo with the **A**IMS) started by forming a consortium with multiple partners, such as PPTs from primary care practices and hospitals, the Faculty of Computer Engineering (HU University of Applied Science), and the Faculty of Social and Behavioural Sciences (Utrecht University).

The home video method, used in four studies in this thesis, was developed specifically around the AIMS for two main reasons. First, the AIMS is a mainly observational measurement tool, and the provision of video footage of their infant by parents, later to be assessed by a PPT, was therefore thought to be feasible. Second, the AIMS is frequently used in clinical practice and research and is a highly valued assessment tool. The first step in the process involved the development of instructions that would enable parents to provide home videos suitable for subsequent assessment by a PPT. Both instructional videos and checklists were designed and tested with the help of parents in two studies: a validation study (n = 48) and a longitudinal pilot study (*n* = 52). Another important part of the process was the creation of a web portal that would guarantee the secure uploading and saving of the home videos. This part of the study was carried out in cooperation with the HU Institute and Research Group of Information Technology, a partner within the GODIVA consortium. Meeting the high standards of the privacy legislation in Europe and combining these requirements with a user-friendly interface for parents and researchers proved to be a challenge.

As stated above, there are several aspects to the development and implementation of a digital innovation. The assessment of motor behaviour from home videos made by parents differs in several ways from a live assessment by a PPT. For example, a live observation concerns a 3-dimensional moving infant while a video shows only a 2-dimensional image of the moving infant. Therefore, it is important to ensure that parents take the videos from the best angle and they have to be guided to video the relevant postures and spontaneous movements of the infant for the PPT's assessment purposes. In this thesis, the validity of the AIMS home-video method compared with a live observation was examined (Chapter 2). Furthermore, the acceptability and user-friendliness of the AIMS home-video method were evaluated from the parent's perspective using questionnaires and interviews (Chapter 3).

Variability in gross motor development

Due to the variability that is observed in the gross motor development of infants, the predictive value of a single observation is low.[11,36,37] If an early delay does not necessarily predict later delay, this has implications for how motor development should be monitored.[12,38] Multiple observations over time are needed to gain a reliable view of the course of development.[39] Where cross-sectional studies focus on developmental differences between infants at various ages, they ignore developmental changes within individuals over time.[40]

In clinical practice, this concept has already taken root. The protocols of well-baby clinics and follow-up programmes for prematurely born infants in specialized clinics are designed in such a way that infants are regularly examined during the first two years of life. Nevertheless, early and sensitive identification of infants at risk who would benefit most from early intervention remains challenging.

For the measurement tools commonly used in the Netherlands to assess gross motor development in infants, it is notable that most norm values are currently based on cross-sectional observations of infants.[22,41] Cross-sectional norm values are suitable for providing information on how an infant is performing at that particular moment in comparison to other infants of the same age but do not provide any information on the trajectory the infant will follow over time.

It has long been recognized that only longitudinal research methods provide true information on developmental outcomes because the passage of time and repeated observations are included in the designs. The research aims can be related to 1) describing motor development (intra-individual change or inter-individual differences in individual change), and 2) explaining motor development (analysing causes of intra-individual change or exploring the impact of factors on the course of motor development).

Over the past four decades, there has been a marked increase in the use of longitudinal studies to examine children's development. Darrah and colleagues conducted a series of studies that demonstrated the intra-individual variability in trajectories of gross motor development in TD children in Canada.[11,12,42] Lately, longitudinal outcomes have also been applied to model condition-specific gross motor growth curves of children with cerebral palsy and children with Down syndrome.[43-45] In the Netherlands, Janssen and colleagues showed that longitudinal motor performance in Dutch preterm infants at 6, 12, and 24 months on the BSID-II was unstable, meaning that the PDI-classification of 85% of the preterm-born infants changed once or more on the three time points measured.[46] However, research on the individual gross motor trajectories of TD Dutch infants is still lacking. Very recent evidence shows that Dutch infants lag in their gross motor development in comparison to Canadian infants.[27,28] This gives an insight into the individual motor trajectories of Dutch infants needed to 1) contribute to the understanding of gross motor development over time, and 2) estimate more accurately whether observed motor behaviour is within the range of normal variation as it develops. The modelling of a gross motor growth curve of TD infants (n = 103) based on the AIMS is described in Chapter 4.

Challenges in understanding gross motor development over time

Factors associated with gross motor development

As stated above, the dynamic systems theory as applied to motor development implies that both child and environmental factors affect gross motor development. The theory emphasizes the role of the development of the nervous system and biomechanical aspects of the growing child within the environment. Ecological theories tend to have a broader scope by focusing more on the context in which the child is developing. According to Bronfenbrenner's ecological theory, development can only be understood by taking into account all systems in which the child develops, both proximal and distal (Figure 1.2). This involves the direct social and physical environment of the child but also, more distally, national health care policies, and cultural values and ideas.[47]

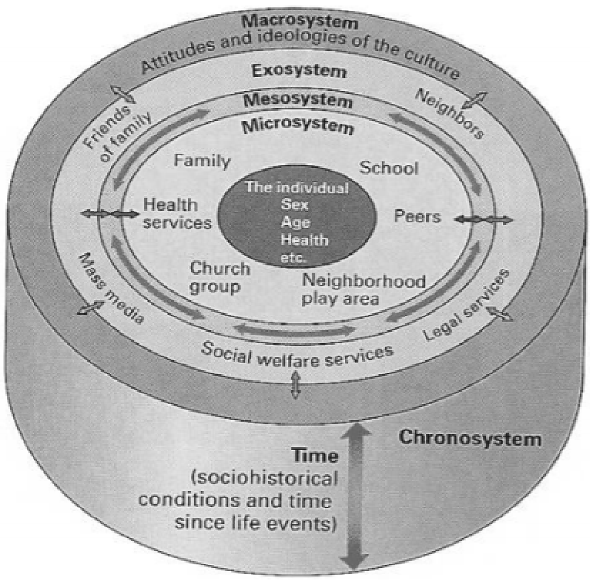

Figure 1.2 Bronfenbrenner's ecological model.
Reprinted with permission of Yingst, N. (2011). Bronfenbrenner ecological systems model. Retrieved June, 13, 2016.

Once the paradigm that development was merely the result of maturation was superseded, research shifted to the investigations of many other factors and their association with gross motor development. The adverse effects of child factors on gross motor development, such as low birth weight and a shortened gestational period, have been studied in depth, particularly in infants born prematurely,[46,48] as have the impact of multiple environmental factors. These studies include, among others, the effects on gross motor development of affordances in the home, the use of toys, sleeping position, and family characteristics among term-born children.[49-52]

Although the number of longitudinal studies is increasing, the majority of studies have evaluated the association of gross motor development with child and/or environmental factors at a single time point during infancy. These studies do not reveal information on whether and if so how such factors have an impact on gross motor development over time.

As previously described, motor development progresses in a non-linear way over time. Research into factors that may influence this development should therefore preferably be studied over multiple time points. The rapid changes during infancy in physical growth, brain maturation, environment, and subsequently motor development, make it plausible that the effect of a factor also changes over time. A systematic review of longitudinal studies on factors associated with gross motor development from birth till independent walking is presented in Chapter 4.

Parental beliefs on motor development

From the viewpoint of early intervention, the impact of potentially modifiable factors on the rate and level achieved of gross motor development is especially interesting. In this context, modifiable factors are factors that are open to change through early intervention to optimize motor development. They are mainly situated in the infant's direct environment, the micro- and mesosystems according to the ecological model of Bronfenbrenner (Figure 1.2).[47] One particularly interesting factor in the microsystem of the infant is the role of the parents. Being highly involved in the daily life of their baby, parents are believed to have a major influence on early motor development.[53,54]

Several studies have convincingly shown that parental beliefs concerning infant development have an important impact on children's development.[55,56] Parental beliefs are defined as ideas, knowledge, values, goals and attitudes.[57-59] However, also other terms are in use, such as parental cognitions and perceptions[58] and ethnotheories.[59]

Harkness and colleagues showed that parental beliefs not only include expectations but also implicit ideas and values that arise from the cultural system a family lives in. The model of ethnotheories shows that the effect of the ideas and thoughts parents have about child development exert an impact on the child's development through caregiving practices.[59] The relation between caregiving practices and infant motor development has been demonstrated in several studies.[60] Differences in parental beliefs and expectations about motor development are present in cross-cultural comparisons of parents with both term- and preterm-born infants.[61,62] Evidence is still lacking, however, on the direct linkage between parental beliefs and a child's developmental course. Research on this subject is challenging due to the many factors that play a role in the progress of motor development and therefore cause "noise." Also, parent-infant interactions and contextual settings are not stable and parental beliefs may change as a result of new experiences.[36] In the model of ethnotheories (Figure 1.3) these are addressed as 'intervening factors'.

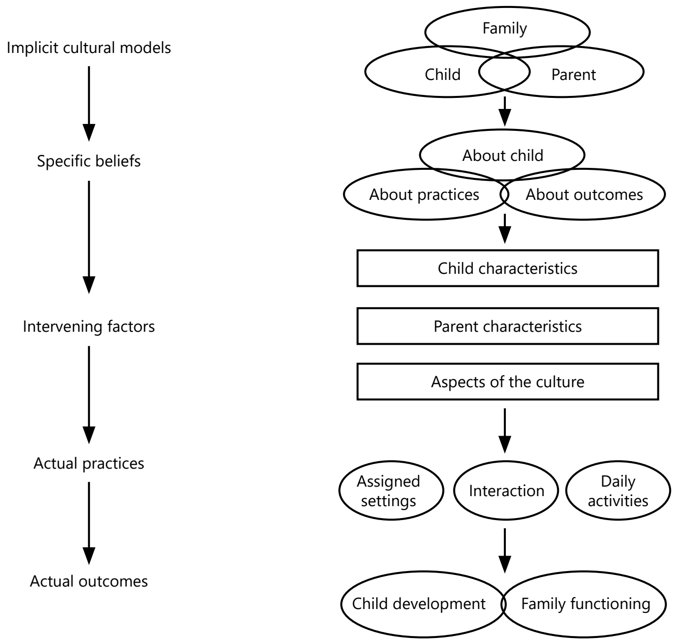

Figure 1.3 Model of ethnotheories, practices, and outcomes.
Reprinted with permission of Harkness S, Super CM, Moscardino U, Rha J-H, Blom M, Huitrón B, et al. Cultural models and developmental agendas: Implications for arousal and self-regulation in early infancy. J Dev Process. 2007;2(1):5–39.[66]

Within the prospective study of this thesis (Chapter 4), we also studied parental beliefs concerning infant gross motor development. To measure parental beliefs, we used the Parental Beliefs on Motor Development questionnaire (PB-MD) that was designed and validated for the Dutch and Israeli populations.[63] The questionnaire was built on the theoretical framework of the developmental niche. This framework conceives culture as the organization of the developmental environment and identifies three subsystems: 1) the physical and social setting in which the child is reared; 2) the customs and practices of childrearing; 3) parental beliefs or ethnotheories.[64]

To assess the parental beliefs on motor development, the PB-MD questionnaire comprises seven statements and four case descriptions, with statements about possible interpretations and ideas on motor development. An example of a case description is: "Noah is a six-month-old boy who is very active and likes to be held in a standing position." The statements that follow reflect a continuum from active stimulation ("Noah should be put in a baby walker"), to a more passive approach ("Parents should not offer Noah the standing position: he might miss the crawling milestone"). Parents rate their agreement to each statement on a 6-point-scale, from 1 (totally disagree) to 6 (totally agree). Furthermore, two open-ended questions are included in the questionnaire about the role that parents have in supporting motor development. Finally, parents are asked to rate the frequency of use of possible information sources on motor development in infancy, such as the Internet, books, friends, and experts.[63] The psychometric properties of the questionnaire were satisfactory to good, with the internal consistency of the subscales between 0.65 and 0.75, and an acceptable to good test-retest reliability. Interviews determined that the convergent validity of the PB-MD was mostly satisfactory.[63]

Cross-cultural evidence revealed that, in the Netherlands, beliefs on rest and regularity prevail above beliefs about the stimulation of motor development among parents.[65,66] As a growing body of evidence points out that Dutch infants seem to develop at a slower pace than in other Western societies,[27,28,41] parental beliefs on motor development are an important factor to consider when studying the progress of infants in their gross motor development.

For health care professionals such as PPTs, parental beliefs on motor development seem to be an under-examined area of interest. Working together with parents in early interventions, knowledge about the ideas parents have regarding (motor) development and child-rearing practices would seem to be an important starting point for collaboration. A prospective longitudinal study on the change in parental beliefs on motor development of Dutch parents (*n* = 78) over time is presented in Chapter 6.

Aims and outline of this thesis

This thesis comprises three main themes: 1) Examining the validity and feasibility of the AIMS home-video method for parents to monitor gross motor development of infants from 1.5 to 19 months; 2) Modelling gross motor trajectories of healthy, TD term-born

Dutch infants from 3.5 to 15.5 months; 3) Identifying factors that are associated with gross motor development in general and, more specific, exploring the parental beliefs about motor development. The research objectives were formulated as follows:

1. To assess the concurrent validity between the AIMS score, based on live observation (established procedure), and the AIMS score, based on home-video observation (new procedure), and the inter- and intra-rater reliability of the AIMS home-video method.
2. To evaluate the feasibility of the AIMS home-video method for parents of healthy, TD infants, born at full term and between the ages of 1.5 to 19 months, from a parent's perspective. What are the expectations and experiences with the AIMS home-video method from the parent's perspective? How do parents evaluate the practical aspects of the home-video method and what are their feelings and thoughts about this new method of assessment?
3. To model gross motor growth curves of healthy, TD term-born infants from 3.5 to 15.5 months based on the AIMS, and investigate patterns within these trajectories.
4. To provide an overview of child and environmental factors associated with gross motor development of infants from birth to independent walking, based on longitudinal studies.
5. To investigate the change in the beliefs on motor development of parents of healthy, TD term-born infants and the associations of the infant's birth order and motor developmental trajectory with that change.

Chapter 2 describes the concurrent validity of the AIMS home-video method when compared to a live observation by a PPT. In this study, 48 parents participated and agreed to make home videos of their infant according to the study instructions. A PPT was simultaneously present in the home to assess gross motor development. The AIMS home-video and live assessments were compared to determine intra-class correlations, mean differences, and smallest detectable changes with the home-video method. **Chapter 3** addresses the feasibility of the home-video method from the perspective of parents. In a pilot study, parents were interviewed and/or asked to complete questionnaires. In **Chapter 4**, the modelling of a gross motor growth curve based on AIMS measurements of 103 healthy, TD term-born Dutch infants aged from 3.5 to 15.5 months is presented. A linear mixed model was applied to model the longitudinal data. Subsequently, cluster analysis was used to identify groups with different trajectories. **Chapter 5** presents a systematic literature review on factors associated with gross motor development of healthy term- and preterm-born infants, from birth to independent walking. This review focused solely on the evidence from longitudinal studies. **Chapter 6** describes the outcomes of the Parental Beliefs on Motor Development questionnaire (PB-MD), administered to parents at their infant's age of 3.5 and 15.5 months. The changes in these beliefs are tested and factors that are hypothesized to be associated with changes are explored. Finally, **Chapter 7** comprises the general discussion of this thesis, including the main findings, considerations, and recommendations for clinical practice and future perspectives.

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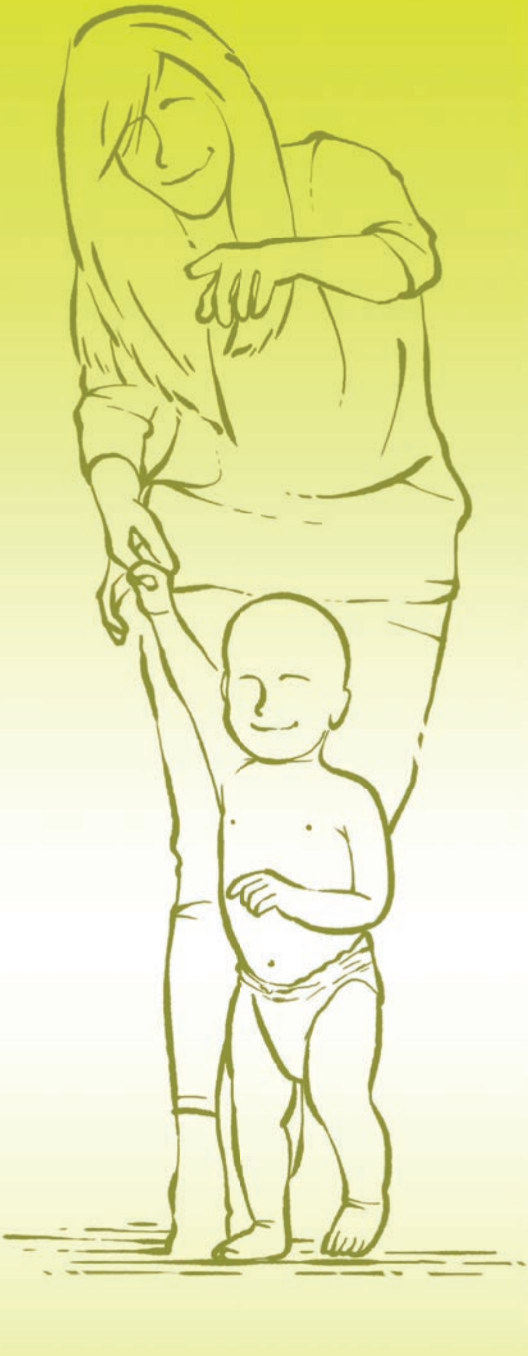
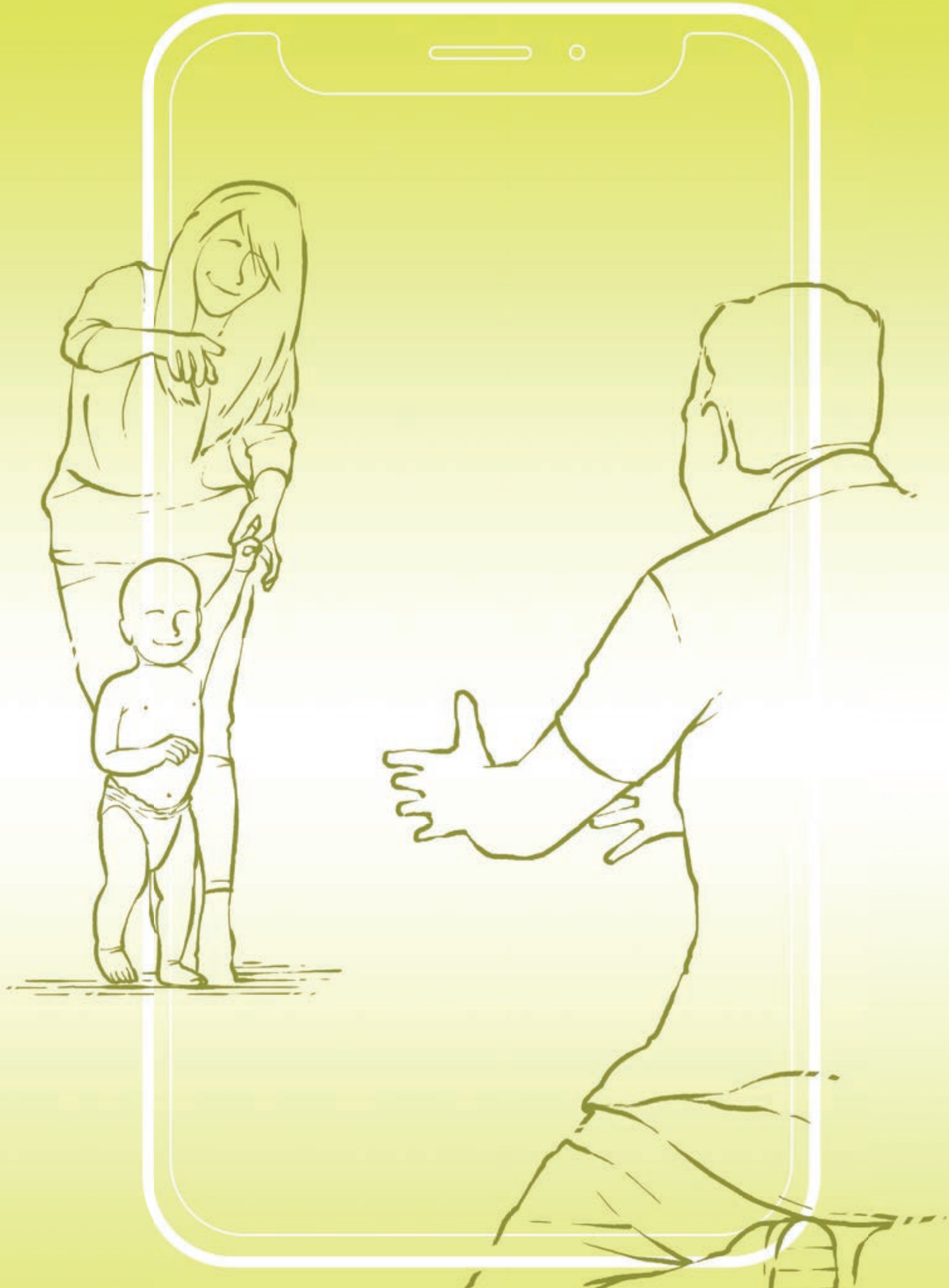


PART I



Chapter 2

Concurrent validity between live and home-video observations using the Alberta Infant Motor Scale

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ABSTRACT

Purpose: Serial assessment of gross motor development of infants at risk is an established procedure in neonatal follow-up clinics. Assessments based on home-video recordings could be a relevant addition.

Methods: In 48 infants (1.5-19 months), the concurrent validity of two applications was examined using the Alberta Infant Motor Scale: 1) a home video made by parents and 2) simultaneous observation on-site by a pediatric physiotherapist. Parents' experiences were explored using a questionnaire.

Results: The Intra-class Correlation Coefficient (ICC) agreement between live and home-video assessment was .99 with a Standard Error of Measurement of 1.41 items. Intra- and interrater reliability: ICCs > 0.99. According to 94% of the parents, recording their infant's movement repertoire was easy to perform.

Conclusion: Assessing the AIMS based on home-video recordings is comparable to assessment by live observation. The video-method is a promising application that can be used with low burden for parents and infants.

INTRODUCTION

Screening gross motor development of infants to detect delays is common practice for a developmental specialist like a pediatric physiotherapist (PPT). The Alberta Infant Motor Scale (AIMS) is a well-known tool to assess gross motor performance in early infancy.[1] However, questions arise about the accuracy of testing an infant on just one occasion.[2-4] The assumption that the sequence and rate of gross motor development is stable within a child might not be correct.[2,5-7] More knowledge on inter- and intra-individual variability of gross motor development in infants is needed,[2,3,8] but longitudinal research is time consuming and testing in an outpatient setting can be burdensome for parents and infant. If the test is pre-planned, there is no guarantee that the state of the infant at that particular moment is good enough to get a valid test score. For these reasons a research project was set up: the **G**ross m**O**tor **D**evelopment of **I**nfants using home-**V**ideo registration with the **A**lberta Infant Motor Scale (GODIVA). Parents are invited to make a structured video of their infants' gross motor repertoire in their home environment.

The reliability and validity of the AIMS are good to excellent.[9] However, applying the AIMS in a home-video setting makes ensuring equivalence a fundamental concern.[10] The main purpose of this study was to assess the concurrent validity between the AIMS score based on live-observation (established procedure) and the AIMS score based on home-video observation (new procedure). We hypothesized that the AIMS score obtained via home-video registration is comparable to the score obtained by live-observation. Examination of the intra- and interrater reliability of the video-method was part of the study. Alongside questions about validity and reliability, feasibility of the video-method for parents were explored.

METHODS

Design

A validation study design was used to determine the concurrent validity of the new and the original method: comparing the gross motor repertoire of infants on the AIMS by a) a home-video made by parents and b) simultaneous observation on site by a PPT. Parents were invited to fill in a digital questionnaire that included questions on the feasibility of the video-method. The study was approved by the Medical Ethical Committee of the University Medical Centre Utrecht, The Netherlands.

Participants

Infants (< 19 months old) and parents were recruited from 01-04-2014 to 31-10-2014 by convenience sampling. Participation was open for parents who were interested in the study (e.g. recruited at birth-centers and well-baby clinics) or with a question on the motor development of their infant (recruited at PPT practices and included before intervention). Parents had to have appropriate understanding of the Dutch language to be included. Infants with known abnormal movement patterns were excluded for this is not the population the AIMS is intended for. When abnormal movement patterns were seen at the video-registration: parents and the family doctor would be informed and the video would be excluded from the study. Parents with a professional background being a physiotherapist were excluded because of their knowledge on motor development. Both parents provided written informed consent.

Assessment tool

Gross motor development was assessed using the AIMS, which was designed to evaluate the gross motor maturity of infants from birth to independent walking.[1] The original norm values were developed based on data from 2202 infants born in Alberta, Canada and recently re-evaluated.[9] The scale contains 58 motor items divided into 4 subscales: prone (21 items), supine (9 items), sitting (12 items), and standing (16 items). Each item is described in detail considering the weight-bearing surface of the body, the posture necessary to achieve the gross motor skill and the antigravity or voluntary movement of the infant. The total raw score can be converted into a percentile rank and/or z-score. The reliability and content validity of the test are described as good.[11,19]

Questionnaire

A digital questionnaire was composed by the researchers and consisted of 25 questions on a 5 point-Likert scale. To characterize the study-sample, questions were included about birthweight and gestational age of the infant. Parents were asked about their age, educational level and knowledge on motor development. Questions on feasibility included technical and operational aspects of the recording.

Procedures

The video-method

The method was developed by experienced PPTs/ researchers. To obtain videos suitable for rating the gross motor performance, decision making for filming was supported by tutorial materials. The method consists of an instruction video and a checklist (Appendix 2.1) for 3 age groups which are adjusted to the motor abilities of the infant: Group 1: 0–5.5 months, Group 2: 5.5–8.5 months, and Group 3: 8.5–19 months. Parents received the instructions that fitted the motor abilities of the infant. To record the video, parents were allowed to use their mobile phone, tablet or video camera. One parent had to record and the other to interact with the infant. When only one parent was present, someone familiar was asked to come over and do the filming. The infant had to be undressed with the exception of a diaper and onesie. Filming was completed when parents thought they had captured the four different postures and movements indicated in the instruction. The home-video ought to be of a maximum length of 30 minutes. The recording was saved at a secured USB device, and stored at our research center according to safety-regulations. Parents received feedback on the motor performance of their child.

The testers

Twelve PPTs, who were familiar with the AIMS, attended two training sessions of three hours led by experts in the field (ICvH, JN). Scoring gross motor performance of infants from videos was practiced and results were discussed using the AIMS administration guidelines. At the end of the training, each tester scored two video-recorded AIMS assessments. In order to be admitted as a tester, one had to obtain a total raw score of each video within a range of ± 2 items compared to the consensus score set in a consensus meeting with four experts (ICvH, JN, EvD, MB) prior to the training. The 2-items range was derived from the acceptable range of the Standard Error of Measurement (SEM) (1–2 items).[1]

Home-video recording and assessment

After inclusion, the tester scheduled an appointment with the parents at home. The parents made a home-video recording while the tester observed the gross motor behavior of the infant simultaneously. The parents were asked to apply minimal infant handling. Motor behavior had to be spontaneous or elicited by presenting toys to the infant. For sake of feasibility questions, the testers were explicitly informed not to help parents making the video or handling the child. The 'gold standard' in this study consists of a live observation where the handling and prompting of the infant is done by a PPT instead of parents. To achieve a better representation of this gold standard, the tester was allowed to do extra observation or handling if necessary after parents completed the video recording.

Afterwards, a second tester rated the motor behavior of the infant on the AIMS based on the home-video recordings. Video assessments were performed at the research center. Both testers were blinded to the AIMS scores of each other to make sure the scores were independent and free from bias. The testers exchanged roles at random during the study.

Inter- and intrarater reliability

All included home-videos were used to evaluate the interrater reliability of the AIMS video- method between three trained testers who assessed the videos again individually. They were blinded with regard to the original score. The intrarater reliability was also evaluated by these three testers. Each tester rescored fifteen videos at random for a second time after a period of at least five weeks.

STATISTICAL METHODS

Concurrent validity AIMS video- and live-observations and reliability

The raw scores were used to determine the degree of agreement between the AIMS scores based on live-observations and the AIMS scores based on the home-video observations. High within-observer agreement is a prerequisite for obtaining valid scores. To analyze concurrent validity, Intra-class Correlation Coefficients$_{agreement}$ (ICCs) for a three-way mixed effects model were used.[12] Given the excellent ICCs of the 'gold standard' (AIMS live) and the appliance of the method in clinical practice, the required level of agreement was set at 0.90.[13] A Bland-Altman (BA) plot with Limits of Agreement was used to visualize the differences between the two measurements.[14] To examine the measurement error in the two scores, the SEM$_{agreement}$ was used and determined to be maximum 2 items, prior to the study.[1] The smallest detectable change (SDC) was calculated from the SEM. To explore the significance of the mean difference, a one-tailed T-test was carried out. To gain more insight in the results, analyses were also conducted on the subscales of the AIMS and on the three different age groups which are described in the first paragraph of the Procedures section. Finally the norm percentile scores[1] were used to explore in how many cases the clinical outcomes on the two assessments would be inconclusive.

Inter- and intrarater reliability AIMS video-method

The $ICC_{agreement}$, the SEM, and the SDC were used to analyze inter- and intrarater reliability of the gross motor assessments with the AIMS video-method. Due to a heterogeneous sample and expecting benefits of rescoring video material, we hypothesized that the $ICC_{agreement}$ for both the interrater and intrarater reliability would be at least as good as the reliability between the live- and video assessments (ICC > 0.90). This applies also to the SEM, < 2 items on the total raw score would be acceptable.[1] Analyses were carried out in the Statistical Package for the Social Sciences 21.0 (IBM SPSS Statistics for Windows, Version 21.0 Armonk, NY, USA).

RESULTS

Twelve testers carried out the assessments (range 3–20). Videos of fifty-two infants, all of good technical quality, were obtained (100%). Four videos (6%) were excluded due to violation of procedures: in one case the infant was wearing clothes during filming, once parents did not use the appropriate instructions during filming because their child was able to roll over but could not show its best motor performance being positioned on a table, the two other videos were not performed on one single day.

The scores of 48 infants (24 males) were compared. The mean birthweight was 3432 grams (range 2500–4365 grams). All infants were at least 37 weeks of gestation at birth. The infants were aged between 1.5–18.5 months. The minimal total raw AIMS score was 3 and the maximum 58.

Table 2.1 Range of age and raw AIMS-scores in three age groups (*n* = 48)

Group	Sample size	Male / Female	Mean age in weeks (*SD*) [Range]	Range AIMS raw scores (live and video)
1	16	6 / 10	16 (5.8) [4.9–25.6]	3–6
2	12	6 / 6	30.3 (6.0) [22.7–42.6]	17–31
3	20	12 / 8	54.2 (10.8) [31.7–78]	32–58
Total	48	24 / 24	35.5 (18.7) [4.9–78]	3–58

Table 2.2 Mean differences raw scores in subscales AIMS (*n* = 48)

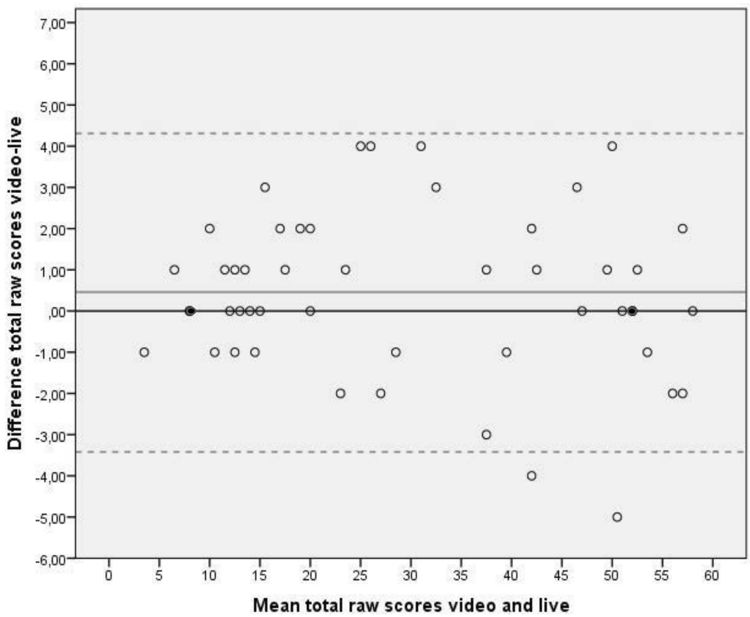
Subscale item AIMS	Sample size	Male / Female	Mean difference Subscale video-live (*SD*)	Range total raw scores (live and video)	Difference in raw score video-live (*SD*)
Prone (21 items)	48	24 / 24	0.13 (0.56)	1–21	0.25 (1.1)
Supine (9 items)	48	24 / 24	0.10 (0.42)	1–9	0.21 (0.85)
Sitting (12 items)	48	24 / 24	-0.02 (0.33)	0–12	-0.04 (0.65)
Standing (16 items)	48	24 / 24	0.02 (0.42)	1–16	0.04 (0.85)
Total (58 items)	48	24 / 24	0.46 (1.98)	3–58	0.46 (1.98)

Concurrent validity AIMS video- and live-observation

To evaluate the agreement between the scores on video-observation and live-observation, a BA plot was used. Figure 2.1 illustrates the differences in AIMS scores between the live-observation and the video-method. The mean difference was 0.46 (*SD* ± 1.98), being not significant (p = .115; 95% CI -.116–1.033) (Table 2.3). In 12 cases there was absolute agreement, in 23 cases the video-observation was rated higher (score difference > 0, mean difference [MD] 2.04 items, min. 1 – max. 4 items), and in 13 cases the live-observation (score difference < 0, MD 1.92 items, min. 1 – max. 5 items). In five cases there were considerable score differences: in four cases 4 items and in one case 5 items score differences between the two observations. Looking at the levels of agreement from a clinical point of view, it was interesting to see in how many cases the clinical outcomes on the two assessments would lead to a different advice to parents. Looking at the percentile ranks and using the *p*5 as cutoff point,[1] in three cases an infant (1, 5, and 8 months old) scored below the *p*5 in one assessment and above the *p*5 in the other assessment.

The $ICC_{agreement}$ between the scores obtained by live- and video-observation was 0.99. The lowest ICC was found in age group 2 (0.89) (Table 2.3). To determine absolute agreement given in items of the test, the $SEM_{agreement}$ was calculated to be 1.41. In age group 3, highest SEM of 1.63 was found while the smallest value of 0.80 was in age group 1. Additionally, the SDC was calculated from the SEM[10] and was 3.88 items. This is the minimal amount of change that must be observed before the change can be considered to exceed the variation and measurement error at the 95% confidence interval (CI).

Table 2.4 presents the ICC, SEM and the SDC of the four subscales of the AIMS. The ICC in supine position is lowest but still good (0.94 item). The SEM is highest in prone position (0.79 item) just like the SDC (2.19 items). This subscale consists of 21 items, the largest amount of the four subscales.



Legend: —— Mean difference total raw scores (0.46, *SD* ± 1.98);
—— Absolute agreement (video score = live score);
------ Limits of agreement (-3.42 +4.33; 95% of scores);
● Represent two cases

Figure 2.1 Bland-Altman plot: video-live (*n* = 48).

Table 2.3 Validity results in three age groups (*n* = 48)

Group	ICC (3 way mixed)	SEM	SDC	MD
1	0.94	0.80	2.20	0.31
2	0.89	1.54	4.27	1.25
3	0.95	1.63	4.50	0.10
Total	0.99	1.41	3.88	0.46 (*SD* ± 1.98)

Abbreviations: ICC, Intraclass Correlation Coefficient; SEM, Standard Error of the Measurement; SDC, Smallest Detectable Change; MD, Mean Difference.

Table 2.4 Validity results in subscales AIMS (*n* = 48)

Subscales AIMS	ICC (3 way mixed)	SEM	SDC
Prone	0.99	0.79	2.19
Supine	0.94	0.59	1.64
Sitting	0.99	0.46	1.28
Standing	0.98	0.60	1.66
Total	0.99	1.41	3.88

Abbreviations: ICC, Intraclass Correlation Coefficient; SEM, Standard Error of the Measurement; SDC, Smallest Detectable Change.

Inter- and intrarater reliability AIMS video-method

An ICCagreement of 0.99 on the total raw scores between three testers indicates a high interrater reliability of the video-method. The average SEM of 0.92 item on the total raw score of the AIMS. The SDC was calculated to be 2.55 items. The intrarater reliability of the video-method was high. ICC on the total raw score was 0.997 (.995–.998). The SEM was 0.96 item and the SDC 2.66 items.

Feasibility

Fifty-one questionnaires were completed by the parents, in 86% by mothers. Almost 75% of the study sample was highly educated. Mean total time needed for going through instructions and filming was reported to be 36.4 minutes (*SD* ± 21.33; range 5–90). 78% of the parents reported that their child demonstrated optimal motor performance or showed new motor behavior. According to 94% of the parents, recording their infant's movement repertoire was easy to perform. 10% of the parents had some doubts about sending a video of their child to professionals. In 96% of the cases, parents reported that making a home video was well to do.

DISCUSSION

The results of this study show high degrees of agreement between an assessment based on a video-registration made by parents and a simultaneous live-assessment of the gross motor repertoire of an infant. The reliability of the video-method itself was evaluated as good, both inter- and intrarater reliability showed high levels of correlation. The conclusions on the feasibility of the video-method for parents are positive.

One of the most important findings in this study is the lack of a systematic difference in the total raw score between the video- and the live-observation scores, nor in the four subscales or in the three age groups. Scores obtained through video assessments were in general slightly higher than the live assessments (+ 0.46 item). In age-group 2, ICC is lowest (0.89 item) while the MD in scores is highest (1.25 item). This finding does not correspond with the ICC's that were found in the reliability study of the original AIMS[1] where correlations were lowest in the youngest and oldest group of infants who performed less items. The lower correlation in age group 2 in the present study, is very likely the result of the smaller sample (*n* = 12). The ICC of the subscale supine is slightly lower (0.94 item) than the other subscales. This might be due to the fact that this subscale consists of only 9 items.

Because there are no guidelines for an acceptable SEM, it has to be defined a priori according to the unit and purpose of the measurement. Prior to the study, a clinically acceptable SEM for the AIMS was set at 1–2 items. A SEM of 1.41 items meets this criterion. In the reliability study of the original AIMS,[1] a SEM was found to be 1.01 on the interrater reliability with two trained testers being present at one occasion, where the primary assessor was administering the test and the other was just observing. The interrater reliability in the present study, combining the live- and video-observation made by parents and rated by different testers resulted in a SEM of 1.41 items.

Because the SEM includes both method variation and between-rater variation, one of the main issues in this study was to establish the source of the error variance when there were considerable differences found between the two scores. Were they due to the between-rater variation or to limitations of the video-method? In two of the six cases when differences are ≥ 4 items, the live-observer rated the infant respectively four and five items higher than the video-observer did. In these two cases this was the result of more handling done by the live-tester after the parents completed filming. However, because the video-assessment scores were in general *higher* than the live-assessments scores, we concluded that differences between the live and video scores in most cases have to be allocated to moderate reliability caused by the involvement of a large number of testers.

With a SDC of 3.88 items, an infant must show a progress ≥ 4 items on the AIMS on the following assessment before it can be seen as a real change (95% CI), not given by measurement errors. In clinical use of the AIMS, we expect this SDC not to be a limitation. It means a progress of, for instance, one item in each of the four subscales. The AIMS has been described to be sensitive to small increments of change over brief periods of time, even like a week.[1] Given the frequency of assessing gross motor development in a clinical setting, it can be expected that the detectable change in a next assessment will lie beyond the measurement error.

In the design of the present study, the method of live-observation and scoring the AIMS was considered to be the 'gold standard' for it is an established procedure in the field of PPTs. By analyzing the data, it was not always possible to establish which score (live or video) was the best representation of the actual gross motor performance of the infant. In some cases the live-observer observed more items but in other cases the live-observer failed to observe items which were present at the home-video. Therefore, the 'gold standard' assumption must be questioned, which means that the outcomes on validity should be interpreted with some caution.

The twelve PPTs from the field who obtained the data, were very diverse in age and years of experience. Making use of this fairly large and heterogeneous group of testers added to the error variance but gave more insight in the potential use of the video method in clinical practice. More research is necessary to establish the added value. However, the fact that agreement was found in this applied research project, is promising.

The high levels of reliability found between and within testers, indicate that the three trained testers can replicate their scores on the AIMS video-method with good accuracy. The SEM and SDC of the video-method are lower (0.92 and 2.66 item) than those of the live- and video-method combined (1.41 and 3.88 item). This is an expected consequence of the involvement of less testers (12 versus three testers) and assessing only the video material. The findings on the home-video method correspond to other reliability studies of the AIMS using video materials.[15-17]

Our study shows that in most cases parents are capable of making suitable videos that can be used to perform a valid assessment of the gross motor behavior of their child. Asking parents to make a video that is used for assessment is quite new; not much is found about this aspect in the literature. The video-method depends partially on an adequate understanding of parents of what and how to film. In recent research papers, there is good evidence of parents being able to provide valid reports on early motor development of their child.[18,19] This implicates that parents have valid ideas about the gross motor development of their child, which might have resulted in an inclusion of 92% of all videos in this present study. However, high educational level of the parents could have positively influenced the quality of the video-recordings. Further research is needed to make clear if the video-method is feasible for parents of different social, ethnic, educational, and economic backgrounds. Also the feasibility of the video-method for parents who have an infant at risk (e.g. prematurity) has to be explored in future research.

This study also raises another important question: What is the best way to observe early gross motor performance? A live-observation is not lasting. Retrospective scoring on the recollection of the observation can be liable to errors. More and more assessors who observe gross motor performance are using video-recordings to improve the objectivity of the observation or test.[20-22] For instance, the agreement between video-recordings and live assessments of the Gross Motor Function Measurement in children with cerebral palsy can be reliably scored using video-recordings.[22] A possible disadvantage however, might be that professionals can only explore the motor performance shown in the video which can be a base for biased or incomplete information or interpretation.[23]

Clinically, the video-method might become a promising addition to the established procedures of monitoring and assessing infants at risk. A key future application of the video- method could lie in longitudinal research projects to develop infant gross motor trajectories. Repeated examiner-administered assessments in longitudinal studies are expensive[3] and can be burdensome for infant and parents. To make this home-video method available for professionals, work must be done to realize a secured web-based design, which enables parents and professionals to interchange videos and feedback.

Another opportunity to use this method is tele consultancy. Parents who live in the countryside and have concerns about the gross motor development of their infant but are not able to visit a hospital or PPT practice can use this home-video method. After uploading their video registration on a safe server, a trained PPT can assess the movement repertoire of the infant and if needed give practical advises or refer to a specialist.

The results of this study indicate that the AIMS home-video method provides reliable and valid measurements that are interchangeable with the live-assessments of the AIMS. However, parents have to follow video procedures to obtain a valid measurement of the gross motor maturity of their child and PPTs have to use the precise descriptions scoring the AIMS. The method allows parents to choose a suitable time for filming, so the infant can show the best motor performance in its own environment. Time and distance become less important barriers. The video is objective evidence of the gross motor performance of an infant and could be retested if needed.

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Author contributions

Marike Boonzaaijer: Conceptualization, Methodology, Investigation, Resources, Data Curation, Formal Analysis, Writing – Original Draft, Project administration. *Ellen van Dam:* Conceptualization, Methodology, Investigation, Resources, Data Curation, Formal Analysis. *Inge-Lot van Haastert:* Conceptualization, Methodology, Writing - Review & Editing. *Jacqueline Nuysink:* Conceptualization, Methodology, Writing - Review & Editing, Funding acquisition, Supervision.

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APPENDIX 2.1
QUESTIONNAIRE VALIDATION STUDY GODIVA

1. What is the code assigned to your child by the GODIVA study? The code contains three numbers.	
2. What is the code assigned to the investigator?	
3. What is your relation to the child?	o Mother o Father/ Partner o Otherwise, namely:
4. What is your date of birth?	
5. What is the date of birth of your partner?	
6. What is the highest level of education you completed?	o No education finished o Primary school o Secondary school/ lower vocational education o Secondary science education/ gymnasium o University/ Higher vocational education o Otherwise, namely:
7. What is the highest level of education your partner completed?	o No education finished o Primary school o Secondary school/ lower vocational education o Secondary science education/ gymnasium o University/ Higher vocational education o Otherwise, namely:
8. My knowledge about motor development is more than average because of my job/study.	o Absolutely o Agree o Neutral o Disagree o Absolutely not
9. I think de motor development of my child is:	o Fast o Above average o Average o Below average o Slow

Questions about the instruction

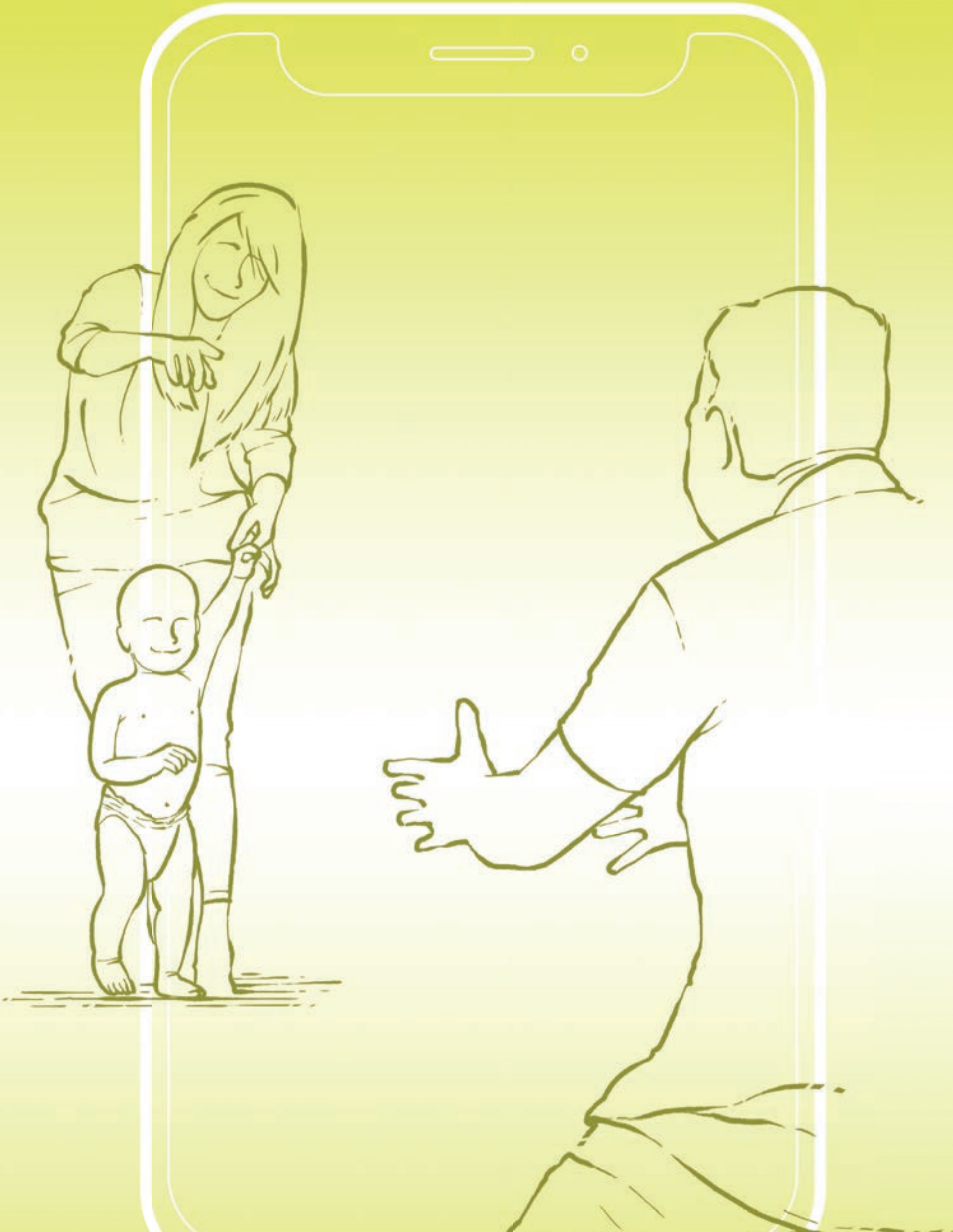
10. The instruction video for making the home video was useful.	o Yes, continue to question 12 o No
11. If you answered 'no': please explain why you feel the instruction video was not useful.	o It was too complex o It lasted too long o The instructions on the location were not feasible o The instructions on prone position were not clear o The instructions on supine position were not clear o The instructions on sitting position were not clear to me o The instructions on standing position were not clear to me o Otherwise, namely:
12. The checklist was an useful addition to the instruction video.	o Absolutely o Agree o Neutral o Disagree o Absolutely not
13. Which checklist and what instruction video did you use for making the home video?	o Instruction video 1: for children o Instruction video 2: o Instruction video 3:

Questions about making the home video

14. My house is a suitable place for capturing the motor skills of my child	o Absolutely o Agree o Neutral o Disagree o Absolutely not
15. Who assisted you during filming?	o Nobody did o My partner o An older child o Otherwise, namely:
16. Do you have the right camera equipment to film?	o Yes o No
17. What was the mood of your child during filming?	o Sleepy, inactive o Calm, inactive o Cheerful, active o Whining, grumpy o Crying, upset, angry o Otherwise, namely:

18. How much time did you spend on preparations and actually making the home-video?	.... minutes
19. My child showed all the motor skills he/she mastered on the home video.	o Yes, even better than that! I saw some new things. → skip question 20 o Yes, I agree → skip question 20 o No, my child has more skills than he/she showed on the home video
20. Which factors had a negative influence on the motor performance of your child?	o Distraction by noises or bustle o Physical discomfort o The mood my child was in o The presence of (strange) persons o No reason, happened by accident o Otherwise, namely:
21. I enjoyed making a home-video of my child.	o Absolutely o Agree o Neutral o Disagree o Absolutely not
22. It was easy to determine if all the required positions and motor skills were captured on the home video.	o Absolutely o Agree o Neutral o Disagree o Absolutely not
23. It was easy to meet the technical criteria for making the home video.	o Absolutely o Agree o Neutral o Disagree o Absolutely not
24. I can imagine the researcher/ Paediatric Physical Therapist is able to assess the motor performance of my child using the home video.	o Absolutely o Agree o Neutral o Disagree o Absolutely not
25. In general, making the home video was well to do.	o Absolutely o Agree o Neutral o Disagree o Absolutely not
26. Did you need help by uploading the home video?	o Yes o No
27. It feels safe to share the home video of my child.	o Absolutely o Agree o Neutral o Disagree o Absolutely not

28. Do you have any remarks or comments on your participation in this study?	



Chapter 3

A home-video method to assess infant gross motor development: Parent perspectives on feasibility

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ABSTRACT

Background: Current use of smartphone cameras by parents create opportunities for longitudinal home-video assessments to monitor infant development. We developed and validated a home-video method for parents, enabling Pediatric Physical Therapists to assess infants' gross motor development with the Alberta Infant Motor Scale (AIMS). The objective of the present study was to investigate the feasibility of this home-video method from the parents' perspective.

Methods: Parents of 59 typically developing infants (0–19 months) were recruited, 45 parents participated in the study. Information about dropout was collected. A sequential mixed methods design was used to examine feasibility, including questionnaires and semi-structured interviews. While the questionnaires inquired after the practical feasibility of the home-video method, the interviews also allowed parents to comment on their feelings and thoughts using the home-video method.

Results: Of 45 participating parents, 34 parents returned both questionnaires and eight parents agreed to an interview. Parent reported effort by the infants was very low: the home-video method is perceived as similar to the normal routine of playing. The parental effort level was acceptable. The main constraint parents reported was time planning. Parents noted it was sometimes difficult to find the right moment to record the infant's motor behavior, that is, when parents were both at home and their baby was in the appropriate state. Technical problems with the web portal, reported by 28% of the parents were also experienced as a constraint. Positive factors mentioned by parents were: the belief that the home videos are valuable for family use, receiving feedback from a professional, the moments of one-on-one attention and interaction with their babies. Moreover, the process of recording the home videos resulted in an increased parental awareness of, and insight into, the gross motor development of their infant.

Conclusion: The AIMS home-video method is feasible for parents of typically developing children. Most constraints are of a practical nature that can be addressed in future applications. Future research is needed to show whether the home-video method is also applicable for parents with an infant at risk of motor development problems.

INTRODUCTION

In recent years, the necessity of multiple testing to monitor infant motor development adequately has been stated in several studies.[1-4] The use of home videos made by parents could be a way to fulfill this need as it reduces the overall burden of traditional testing on infants and parents. The availability of the Internet and digital cameras, important conditions, seem to have been met, for 98.7% of persons between 25–45 years use a smartphone in the Netherlands (Statline, 2018).[5]

For this reason, we developed and validated a home-video method which enables professionals to evaluate gross motor performance with the Alberta Infant Motor Scale (AIMS),[6] a valid and reliable assessment tool for infants (0–19 months).[7-10] An important advantage of this assessment tool is that it evaluates spontaneous motor behavior and requires minimal handling. The home-video method allows parents to record their child's motor behavior at home and at a convenient time, which increases the chance that the infant will show optimal motor performance.[6] Parents make a home video of their baby, guided by instructions. Then, they can upload the videos from their smartphone or camera through a computer to a web application which was specifically designed for this purpose. The videos are stored after encryption, with individual encryption keys assigned to each participant. The server has been tested successfully with a high-level security scan by both the institutional security office and an independent outside security office. A Pediatric Physical Therapist (PPT) can then observe the videos and assess the infants' gross motor development with the AIMS. Unlike a visit to an outpatient clinic, time and geographical distance are no longer barriers.[9] Figure 3.1 provides a detailed description of the home-video method.

The instruction The method comes with three instruction videos and three checklists to guide the caregivers/parents. These roughly fit three age groups within the AIMS (0–5.5 months, 5.5–8.5 months and 8.5–19 months). The checklists (Additional File 3.3) support parents during filming to determine whether they captured the entire motor repertoire of their baby.

Recording the home video The home video can be recorded with a smartphone, tablet or camera. The maximum time frame is 30 minutes but 10–15 minutes is sufficient.

Uploading the home video To make safe uploading of video material possible, a web portal was developed. After verifying credentials, parents have direct access to a secure streaming server to upload their home videos.

Feedback Within two weeks, caregivers/parents receive feedback on the motor development of their baby by email. If desired, caregivers/parents can receive feedback by telephone as well. In case the infant scores below the cut-off point (5th percentile) parents are contacted by telephone and if necessary the family practitioner will be informed.

Figure 3.1 The AIMS home-video method.

Lately, the use of home videos made by parents to assess or evaluate development has been the subject of several studies.[11-15] Libertus et al. successfully used Skype and FaceTime to assess infants' early motor skills.[13] Using this method, the digital live connection with parents provided the opportunity to guide parents during the assessment. Although the study stated that using parents in the role of experimenter could lead to increased assessment variability, overall the conclusions on the feasibility for parents were positive. A pilot study by Ricci et al. on the feasibility of filming the General Movements Assessment (GMA is a 3-minute video of the infant's spontaneous movements in supine position) by parents at home after Neonatal Intensive Care Unit discharge showed a less positive outcome.[14] During this pilot, parents experienced major problems recording and sending accurate videos. Therefore, the clinical feasibility of providing adequate home videos made by parents could not be determined. Recently, Spittle et al. launched the Baby Moves Application for parents to record GMA.[12] The usability of the app and the engagement of 451 parents was evaluated by Kwong.[15] This population-based study included 226 infants born extremely premature or with an extremely low birthweight and a control group of 225 term born infants. Overall, positive results on the usability of the application are reported, most parents were able to successfully capture their infant's movements with the app. All studies carried out so far focus on the practical feasibility of the use of home videos in assessments.

The uniqueness of the AIMS home-video method lies in the fact that parents have a leading role in executing the first part of the assessment, capturing gross motor performance. Apart from the instructions, parents do this on their own. Because most e-Health innovations do not make it to implementation in clinical practice,[16] the feasibility of the home-video method for parents needs to be considered carefully.[17,18] It is important to gain insight into (1) how parents evaluate the practical aspects of the home-video method, and (2) the new role they have in the assessment.[17,19] Examining these aspects with parents of typically-developing (TD) infants is a first step in our ongoing research project. Parents of infants at risk, using the home-video method, are the ultimate target population.

Thus, the overall objective of this study was to evaluate the feasibility of the AIMS home-video method for parents of TD infants, born at full term and between the ages of 1.5 to 19 months, from the parents' perspective. In this study, feasibility was defined according to Karsh as 'the extent to which an innovation can be successfully used or carried out within a given setting'.[18] According to this construct, we formulated two research questions: (1) how do parents evaluate the practical aspects of the home-video method? and (2) how do parents feel and what do they think about this new method of assessment?

METHODS

Study design

Because the present study not only focused on the process of the recording but also on parents' experiences in this specific context, a prospective mixed methods design was chosen.[20] In a mixed methods design, both numeric data and textual information are used, which can be gathered simultaneously or in a sequential manner.[20-22] In the present study, a sequential design was used because of the longitudinal nature of the pilot study[23] (Figure 3.2). To evaluate the practical aspects of feasibility, questionnaires were used.[18,19,24] To gather more in-depth information on how parents evaluated their new role and to reveal barriers and positive factors, both open-ended questions in the questionnaires and semi-structured interviews were used to collect qualitative data. The quantitative and qualitative data were analyzed separately, and results were integrated while interpreting the findings.

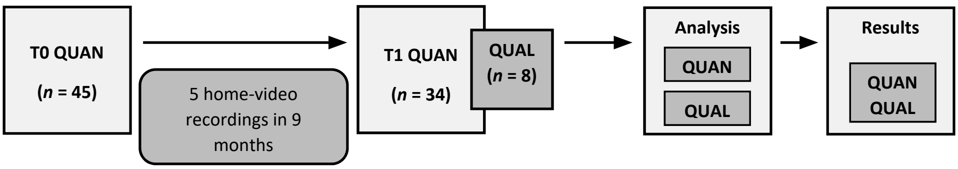

Figure 3.2 Model of mixed methods design.

Setting and participants

Study participants were parents of full-term-born TD infants (1.5–16.5 months) who had participated in a pilot study on longitudinal gross motor trajectories (n = 45) in the Netherlands. Parents were instructed to make five home videos of their child with a two-month interval between each video. Two cohorts of infants were included in the study, starting simultaneously. The first cohort comprised 18 infants who started at the age of 1.5 months and were subsequently recorded on video at 3.5, 5.5, 7.5 and 9.5 months. Infants in the second cohort (n = 27) were recorded by parents at the ages of 8.5, 10.5, 12.5, 14.5 and 16.5 months. The time frame for making each video was set at exactly two weeks. During the study, parents received reminders by e-mail of when to record a video.

The recruitment of parents took place by word of mouth, at social media, day care centers and well-baby clinics by convenience sampling from June 2015 to July 2016.

Because of the digital nature, there were no geographical boundaries to participation. Parents expecting or having a full-term-born TD infant and who understood the Dutch language were eligible to enter the study. A subset of eight parents from the study sample was selected for interviewing through a purposive sampling approach to ensure variation in parental and child characteristics, namely age, sex and education level of the parent, birth rank and motor development of the infant. The aim was not to generalize but to obtain a wide view on parental experiences regarding the home-video method.

Questionnaires and interviews

Online questionnaires were used to enquire into parents' expectations (T0, before the first video moment) and actual participation (T1, after the last video moment, see Figure 3.2) regarding the home-video method. The questionnaires, developed by the researchers, consisted of 21 questions at T0 and 24 questions at T1 (Additional File 3.1). Questions were included on parent and child characteristics, and on the usability of the home-video method and the web portal. A 5-point Likert's scale was used (1 = strongly agree it is easy to perform; 2 = agree it is easy to perform; 3 = neutral; 4 = disagree it is easy to perform; 5 = strongly disagree it is easy to perform). A priori, acceptable outcomes in terms of feasibility were set at < 3.

To quantify the expected and experienced effort level for parent and infant (parent-reported), a 10-point scale was used at T0 and T1 (0 = no effort; 10 = a lot of effort).

To obtain information on the children's longitudinal motor trajectories, Question 21 (T0) and Questions 20–23 (T1) were added to the questionnaires but not included in the current analyses.

A topic list (Additional File 3.2) provided the basis for the semi-structured interviews. The interviews with the parents, conducted by the first author, took place at home and lasted 30 to 45 minutes. One respondent preferred to do the interview at work. The interviews were planned after the parent filled out the second questionnaire (T1), recorded on audiotape and transcribed verbatim.

Ethical aspects

The study was approved by the Medical Ethical Board of the University Medical Centre Utrecht (METC/UMCU) reference nr.14-399/C, and both parents gave written informed consent. Additional written consent was obtained for the interviews.

Data analysis

I Quantitative analysis

The mean and standard deviation on single items of the questionnaires (T0 and T1) were calculated. Paired samples t-tests and Wilcoxon signed-rank tests were applied to detect changes in expectations and experiences of parents between T0 and T1. Only parents who filled in both questionnaires were included in the analyses (*n* = 34). Statistical analysis was carried out with IBM Statistical Package for the Social Sciences 21.0 (IBM SPSS Statistics for Windows, Version 21.0 Armonk, NY USA).

II Qualitative analysis

To analyze the data from the interviews and from open-ended questions in the questionnaires, a thematic analysis with a general approach was used, guided by the research questions.[25] After familiarization with the data by reading the transcripts, relevant fragments were coded independently by two researchers (CdB, MB) using MaxQda 10 software.[26] Codings were discussed until consensus was reached. During this process, the codes were categorized into a structured code tree. Emerging themes were identified by constant comparison of codes and text fragments.[27] Although the main focus of the analysis was deductive, based on the topic list, in each phase there was room for inductive elements.[28] The main themes and subthemes that were identified were linked if possible and an overarching interpretation achieved.

RESULTS

Although 59 parents provided informed consent, 45 participated in the pilot study. Parents who did not send in home videos were approached by telephone to inquire about the reasons for not participating. Reasons for dropping out were: 1) the baby was unexpectedly born prematurely or pathology became evident shortly after birth (*n* = 2); 2) parents reported that in retrospect they were too busy to participate (*n* = 11); 3) frequency of filming was too high (*n* = 1). Participating parents were residents of 8 of the 13 different provinces in the Netherlands. In total, 45 questionnaires were returned before the start of the study. Following the period of recording the five home videos, 34 surveys were returned (T1; response rate 76%). Table 3.1 shows the characteristics of participating parents at T0. From this group, 10 parents were approached for an interview. In two cases, parents were unable to schedule an appointment in the allocated period; the other eight parents agreed to an interview.

Table 3.1 Infant, parent and home-video characteristics

Infants (*n* = 45)	
Female (%)	44
Gestational age in weeks (*M*, *SD*)	39.27 (1.45)
Birthweight in grams (*M*, *SD*)	3432.7 (504.1)
Birth rank (%)	1st (64)
	2nd (30)
	3rd (6)
Parents (*n* = 45)	
Mother/Father (%)	42 (93)/3 (7)
Age (yr, %)	25–30 (24)
	31–35 (56)
	36–40 (13)
	41–45 (7)
Education (%)	Medium (7)
	High (93)
Home videos	
Total number of recordings	185
Number of recordings per infant (*Mdn*, Range)	4 (1–5)
Device used (%)	Smartphone (60.6)
	Digital camera (27.3)
	Tablet (6.1)
	Other (6.0)

Legend: *M* = Mean, *SD* = Standard deviation, *Mdn* = Median.

After analyzing both quantitative and qualitative outcomes, the final thematic framework comprised two main themes: 1) feasibility of the home-video method, in which we combined both quantitative and qualitative data to gain insight into the extent that parents can carry out the home-video method successfully, and 2) parents' feelings and thoughts that accompany the use of the home-video method. These results were mainly inductive qualitative outcomes.

The findings are structured according to the process of making the home video: reading the instructions, planning when to make the recording, recording the home video, uploading the home video, and receiving feedback. First, the quantitative data are presented; next, the qualitative data are used to set the context and to clarify the quantitative findings. In Table 3.2, the quantitative outcomes are shown and in Figure 3.3 the qualitative findings are summarized and visualized.

Table 3.2 Quantitative results of Expectations (T0) and Experiences (T1) of parents applying the home-video method

	T0 Expectations (n = 34)	T1 Experiences (n = 34)	T0–T1
Effort of home-video method	*M* (*SD*)	*M* (*SD*)	Paired t-test ($t(df)$, p)
(0 = no effort, 10 = a lot of effort)			
Parental effort	3.72 (1.67)	4.00 (2.33)	$t(33) = -0.545$, $p = 0.590$
Infant effort (parent-reported)	1.97 (1.74)	1.55 (1.48)	$t(33) = 1.046$, $p = 0.304$
Practical aspects of the home-video method	*M* (*SD*)	*M* (*SD*)	Wilcoxon's *SRT* (z, p)
(1 = strongly agree easy to perform, 5 = strongly disagree easy to perform)			
Technical aspects of recording	1.83 (0.54)	2.10 (0.86)	$z = -1{,}99$, $p =$ **0.046**
Positioning the infant	1.72 (0.53)	1.69 (0.60)	$z = -0.26$, $p = 0.796$
Prompting movements	2.04 (0.64)	2.07 (0.81)	$z = -0.23$, $p = 0.819$
Uploading	2.0 (0.89)	3.38 (1.18)	$z = -4.08$, $p <$ **0.001**
Finding a convenient moment		3.21 (1.01)	
A 2-week window is sufficient		2.47 (1.05)	
Instruction videos were clear		2.06 (0.74)	
Checklists were clear		1.56 (0.61)	
Feedback no reason for concern[1]		1.93 (1.26)	

Legend: M = mean; SD = standard deviation; df = degrees of freedom; t = t-value paired samples t-test; z = z-value Wilcoxon *SRT*; [1]Outcome item 'Feedback no reason for concern' was recoded.

3

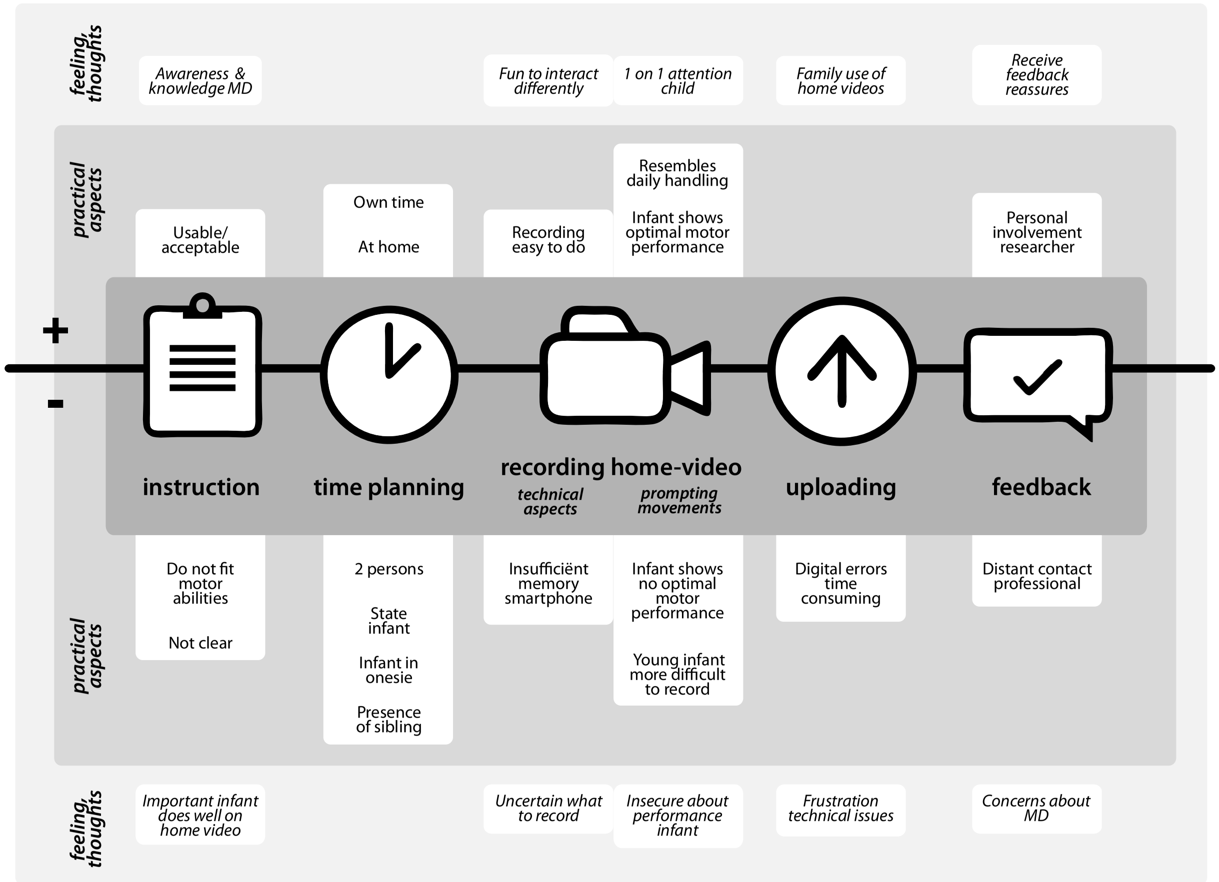

Figure 3.3 Qualitative results of parents' perspectives on the feasibility of the AIMS home-video method regarding 1) the practical aspects and 2) the feelings and thoughts.

I Feasibility of the home-video method according to parents: Practical aspects

Expected and experienced effort for parents and infants

The quantitative data showed that the expected and experienced efforts of parents applying the home-video method were similar. Qualitative data revealed that parents who appraised the effort higher than expected primarily attributed this to technical issues during the uploading: '*I didn't think recording the video was very burdensome. Besides, it was fun to do. But, because of the technical issues uploading the video, it took much longer than expected and that made it somewhat frustrating*' (123, mother).

Both the expected (M = 1.97, SD = 1.74) and experienced (M = 1.55, SD = 1.48) parent-reported effort of the home-video method for the infant were rated low, and not statistically different for T0 and T1. Parents highlighted this by stating they were primarily recording their baby's spontaneous movements: '*The video and the small exercises were no effort for him, I think he actually enjoyed it*' (118, mother). In some cases, the infant was not in the right state, which made the recording a bit more demanding: '*For as far I could see, it was no burden on my daughter. Sometimes, she was not in the mood but the exercises were not annoying. Besides, most of the time we were recording her spontaneous movements*' (104, mother).

3

Instructions

The parents rated the usability of the instructional videos as good. Furthermore, they described the checklists as very usable and clear (M = 1.56, SD = 0.61) (Table 3.2). The qualitative data supported these findings. Most parents reported viewing the instruction video prior to the recording and using the checklist during the recording: '*The checklist was very handy, we had that at hand every time to see: did she show just about everything? It was sort of a guidebook. O.K., we put her down and we have to make sure she does all these items. I also thought, in terms of design, it looked really clear and gave explicit instructions*' (145, mother). In a few cases, parents encountered some difficulties applying the checklists because they felt that none of the checklists fitted their infants' motor abilities adequately at that time: '*The first checklist, well, I felt like: this is too easy, he can do all this already. Checklist 1 was far too simple and he couldn't do much of checklist 2*' (118, mother).

Time planning

Quantitative data showed that parents thought planning the time to record the home video to be an impediment (*M* = 3.21, *SD* = 1.01). The 2-week window in which parents could record the video was not always sufficient (*M* = 2.47, *SD* = 1.05). These outcomes correspond with the qualitative data where time planning was expressed by a majority of parents as being the main barrier to recording the home video. Parents also mentioned other factors which interplayed with this main barrier. The necessary presence of two persons to record the home video made time planning more complicated. '*I found it quite hard because we both spent a lot of time at home with her, but not much time with all three of us*' (118, mother). One mother explained how the recording of a very young infant could also lead to planning problems: '*I also breastfeed and certainly in the beginning that takes such a long time so then it's often when they're awake you are busy feeding and afterwards they need a change, and those kind of things. Putting them on their tummy and exercising them was not an immediate priority*' (144, mother of twins).

Also, the fact that parents preferred to choose a moment when the infant was in the right state for recording added up to quite a complex puzzle in today's dynamic family life. '*Sometimes it was just difficult timing, you think oh yes now, but then they are tired and then, you really want them to show their best, and then you think: no, they are too tired to do it now*' (114, mother).

Finally, the presence of an older brother or sister in the toddler or preschool age, could pose a dilemma: '*Besides, we have another 5-year-old daughter who we didn't want to have around at that moment because she wants the attention as well. We really needed to look for occasions when she wasn't at home*' (124, mother).

Parents also experienced favorable aspects of the home-video method, such as being able to video the infant at home in their own time without a professional coming over to assess the infant's gross motor development. '*Would I have preferred a researcher coming over here for each video moment? On the one hand, then you make the appointment and then it is set, yes? But then you're stuck with it. This way, I could plan it in my own time. So, that's a big advantage of doing the recording by yourself*' (136, mother).

A father puts it like this: '*It is of course very accessible, you don't have to leave, nobody has to come to your house and you can record a video and get a reaction to that. So I think it can only be more convenient*' (152, father).

The home appeared not to be the only suitable place for recording the videos. In multiple cases, infants were recorded during a visit or stay with the grandparents. Also, during holiday seasons, some parents sent home-video material from camping sites, apartments and cottages from all parts of the world. '*We went on a holiday and made the videos, and once we recorded the video at my parents' house, so we did film her at diverse locations. With such a small baby that is no problem, of course*' (136, mother).

Technical aspects of recording

In advance (T0), parents did not expect (M = 1.83, SD = 0.54) the technical aspects of the recording (i.e. camera position, light and distance) to become a problem. At T1 (M = 2.0, SD = 0.86), the experience was rated not much but still statistically significantly higher in difficulty (Z = -1.99, p = 0.046) (Table 3.2). The opinion of most parents can be gathered under this parent's expression: '*The recording itself was not hard to do; I do it every day!*' (141, mother). However, due to the daily use of the smartphone as a camera, some parents already had a lot of photo and video files stored on their smartphone. This might explain the significant negative change in the experiences parents had regarding the technical aspects: '*After a few videos, the memory card in my smartphone was full. So I had to upload and remove photos, which takes time. After that I'm able to continue recording, in the hope my baby still wants to cooperate*' (114, mother).

Positioning the infant and prompting the movements

Parents found it easy to position their child in accordance with the instructions (M = 1.72, SD = 0.53 expected and M = 1.69, SD = 0.60 experienced: Table 3.2). This can be understood from the qualitative data too, where parents explained that it mostly resembles daily handling: '*She did what she is always doing, only now with a bit more facilitation and a camera present*' (152, father).

Parents also rated the prompting of specific movements as feasible to perform (M = 2.04, SD = 0.64 expected and M = 2.07, SD = 0.81 experienced: Table 3.2). A mother expressed in the interviews: '*You really prompt her, yes. She has now reached out with her right arm and then you try to get her to reach with her left arm also. So that's what I really enjoyed*' (145, mother).

Although most infants were recorded at a convenient time and in the right state, some parents noted that their infant did not show optimal motor performance during recording. In the questionnaire, 23% of parents indicated that their child did not show optimal motor performance in the final home video. Reasons for this were 1) the state of the infant, 2) the infant was distracted by the camera and 3) by coincidence. This could lead to some frustration for both parent and child: '*It was hard to find a moment* *he was in the right mood... so sometimes he got frustrated for not showing things he normally would show and we were waiting for him to show that behavior*' (123, mother). However, 77% of parents stated their child did show optimal motor performance or even showed new motor abilities during the recording.

Uploading the home video

In advance, parents did not expect that uploading the home videos to the web portal would lead to any obstacles (M = 2.00, SD = 0.89). However, afterwards this theme demonstrated a significant negative change (M = 3.38, SD = 1.18, $p < 0.001$). Due to instability of the software during the pilot, the web portal was not always functioning properly which made uploading more time-consuming. Approximately, 28% of parents encountered these difficulties. Parents also reported this as a factor that increased the overall effort they experienced during the pilot. Where mothers were most involved in the study, fathers played an important role in dealing with the digital problems. '*I kept aloof from that [uploading home videos], I am not that into transferring videos onto the computer, so that was my husband's thing. I was into the recording and telling him what we had to do and he mainly did the technical part*' (136, mother).

Receiving feedback

In the questionnaires, most parents reported that the feedback on the motor development of their child gave no cause for concern (M = 1.93, SD = 1.26) (Table 3.2). Furthermore, some parents reported that the feedback and access to an expert on motor development they could turn to with questions was an agreeable aspect of participating. '*And if something goes wrong, he lags behind or there is a handicap, that you know it in time. That there are professionals monitoring your baby who can intervene in time. So you don't just find out at the age of 4 that he can't throw a ball*' (114, mother). In this context, the feedback was mentioned as an important motivator to stay involved in the study.

One parent thought the feedback was a less important part of the process. For her, seeing her baby perform was the most enjoyable element: '*The feedback was nice to see but the fun part was the moment that you record her and see her doing it*' (145, mother).

II Parental perspective on the new role: Feelings and thoughts

In addition to the perspective parents gave on the practical aspects of feasibility, they also expressed their 'feelings and thoughts' which accompanied their new role in applying the AIMS home-video method. Parents expressed both negative and positive feelings and thoughts. In Figure 3.3, these results are displayed in the outer part of the model.

During recording, some parents experienced insecurity about the motor development of their child. Also, some of them reported insecurities about whether they had recorded the movements and postures as intended. Especially when recording for the first time, they expressed questions about the duration of the recording and how long they should keep on facilitating: '*You are just not sure if you did the recording the right way, so I just went ahead and made the video but still I wasn't certain*' (118, mother).

The qualitative data showed that a few parents, whose children scored below average on one or more occasion, did experience some concerns when they received feedback: '*At the start, I found it a bit difficult to see that T. scored quite low, but that was a result of my insecurity as a mother*' (106, mother).

Almost all parents expressed it was important their child would show the best on the home video: '*At that moment, I wanted him to show the good things, yes I felt quite strong about that. After all, you would get feedback on it and it was about his development. You knew he already was able to do some things but when he was tired, he didn't show it that well*' (114, mother). Some parents even considered making a new recording because they were not satisfied with the first. However, parents refrained from this because of time constraints.

Many parents reported that, despite the effort involved, they did enjoy the individual attention and time spent with their baby: '*And somehow, with your firstborn you probably have it [one-on-one attention] more. She is my second and I almost felt like I wanted to give her this attention to her motor development*' (145, mother).

The active involvement of parents in recording the home video appeared to have some side effects triggered by the fact that parents interacted with their baby in a different way. By looking at the instruction video and the checklists, several parents reported they gained knowledge about, and became more aware of, their baby's motor development: '*So I did notice, especially in the beginning, that suddenly you start realizing what she is doing. You really start very focused observing*' (145, mother). In one case, parents were alarmed by what they observed in the instruction videos: '*By looking at the instruction videos, we realized that our son lagged behind in his motor development, so we contacted a pediatric physical therapist*' (121, mother).

Some parents also acquired new insights in how to optimize motor development: '*Yes, well also regarding tummy time, we found out that the baby enjoyed to move around on a larger surface. Because we saw the effect it had, we did it more often*' (114, mother).

For the participating parents, who all have TD infants, the main encouragement to participate was to obtain valuable home-video material which captured the motor development of their baby over a period of time. Another key to compliance was the feedback on their infants' motor development. Parents found the extra developmental monitoring of their infant both reassuring and interesting.

DISCUSSION

The present study explored the experiences of parents in using a home-video method to assess their infants' gross motor development. Overall, parents were positive about the practical feasibility of the home-video method. They reported that the recordings were easy to do and that the handling of the baby was mostly as in daily routines. Several barriers were identified in this study. The main barrier reported was time planning. A second barrier concerned technical problems with the web portal, which sometimes made uploading the home videos time consuming. According to parents, positive factors of this home-video method were (1) that the home videos were valuable for family use, (2) that receiving feedback from a professional about infants' motor development was welcome, and (3) that it was fun to interact with their babies in a different way and to have a moment of one-on-one-attention. Moreover, the instructions and home-video recording resulted in an increased parental awareness of, and insight into, the gross motor development of their infants. The feelings and thoughts parents expressed about their new role were both positive and negative. In some cases, parents expressed their uncertainty about the motor performance of their child or about the video recordings. Parents also reported joyful feelings about the interaction they had with their baby while making the home videos. In addition, most parents appreciated the feedback on the motor development of their child which they found reassuring.

For future application, it is important to address all barriers identified in this study.[19] Time planning is mentioned most explicitly: parents were hard pressed to find a moment when they both were at home and their baby was in the proper state to show optimal motor behavior. During the study, some parents found a solution to the logistics: by positioning their phone on the table or floor, they managed to record and handle their infant at the same time.

From the results, we can conclude that a functional and user-friendly digital application is an absolute prerequisite for successful implementation of this method. This is exactly in line with the conclusions of Ricci et al.[14] The main barrier they described was the use of an encrypted server with very high protection levels, obligated because the home videos were considered to be personal health information. In this study, the server was security tested and found to be compliant to relevant laws (NEN 7510/7512/7513 norms). The encryption of data while uploading is important to ensure safety but as a consequence the uploading was sometimes time consuming. This was also the case for the assessors while downloading and decrypting the video data. Both aspects limit feasibility and should be addressed. A satisfactory compromise between functionality and safety in the development of health care applications seems an important step towards successful implementation in practice.

In addition, in the development and use of digital communication means, the privacy of parents and infants is considered to be very important.[29] In our study, privacy issues did not emerge as a significant theme. Perhaps digital privacy is not an important issue for all parents. Ricci et al.[14] reported that, because of the problems uploading the videos, many parents offered to share the home videos on open platforms like Facebook or WhatsApp. In our study we had similar experiences. This might also be in line with the findings of Hassol and colleagues, who reported that only a minority of patients was concerned about the privacy of their electronic health care record.[30] However, a self-selection bias may have occurred in the privacy aspect. Parents with explicit ideas on privacy regarding video material of their child may have decided not to participate in the present study from the start.

Libertus and Violi, who used Skype as a means of collecting developmental data, suggested that access to the Internet and digital equipment could also be a constraint for parents' participation in these kind of research projects.[13] In our homogeneous sample, all parents had access to the Internet and a smartphone. According to Statistics Netherlands, over 98% of persons aged 25–45 years have access to the internet and almost 95% own a smartphone.[5] These high percentages lead us to believe this aspect unlikely to be a limiting factor for participation in our study.

Only a few studies describe the feasibility of digital screening methods for parents at home.[11,13-15,24] Besides, every method has its own specific features which affect parental experiences and thus feasibility in different ways. The evaluation of the usability of the Baby Moves app shows that most parents successfully used the app to record their baby's movements.[15] However, because the AIMS home-video method is more demanding for parents, it is questionable whether these results can be applied to the AIMS home-video method. Our positive findings on the feasibility of the AIMS home-video method are more comparable to a study on a video-based evaluation tool for children with Rett syndrome.[11] In this study, outcomes on feasibility were positive, despite the fact that parents had to follow quite extensive instructions to record multiple abilities and interactions. Furthermore, these authors reported benefits from recording the child in a familiar setting. We think this aspect also applies to a large extent to the AIMS home-video method. On most home videos, infants' state was suitable for testing. When assessing motor development from the recordings, it was seen that the infant didn't have to adapt to a new environment, strange people or a set appointment time, which is the case when the infant is seen in a PPT practice or hospital outpatient clinic.

Although some parents reported that their child did not always show optimal motor performance on the home videos, we speculate that this might be overstated. The importance parents placed on their child's showing optimal motor performance on the home video might sometimes have resulted in a more negative perception of the child's performance. For example, if an infant had shown rolling over from supine to prone for the first time just before the recordings, it is quite likely not to be shown in the home video, and parents could feel disappointed about this. For a professional assessing the home video, not seeing the infant rolling over would not necessary influence the validity of the assessment; rolling over might just not yet be in the infant's motor repertoire.

An important finding of this study is the teaching effect the AIMS home-video method potentially has. The method requires active parental involvement which can lead to a better understanding of the infant's motor development.[31,32] Parents with an infant at risk for delay might especially benefit from this knowledge. It might help them to become 'their child's expert' even more and as such improve equality in shared decision-making between parents and professionals.[33]

Strengths

This study is the first that not only reports outcomes on practical feasibility of home video assessments but also attempts to grasp the feelings and thoughts of parents. Parents are the most important stakeholders in the home-video method and their experiences have to be acknowledged for successful implementation. The mixed methods design, a combination of questionnaires and interviews, provided rich information about the experiences of parents. The main outcomes of both qualitative and quantitative data reinforced each other and were thus complementary. The interviews clarified and illustrated the quantitative findings.[22] The thematic analysis with a combined approach, both deductive and inductive, brought forth important new insights in parents' feelings and thoughts regarding the home-video method. Another strength concerns the longitudinal nature of the study, which allowed parents to report on multiple experiences with the recording of their child, instead of a one-time exposure. Because of this design, it was also possible to inquire after the expectations of parents before the start of the study.

Limitations

Our study is subject to the following limitations. The advanced educational level of the majority of participating parents limits the generalizability of the results. The checklists do demand some literacy and might therefore not fit parents who are less educated. On the other hand, the additional instruction videos could partially solve this barrier. In the population-based study of Kwong et al., it became evident that families of lower socio-economic status who used the Baby Moves app were less likely to return scorable videos.[15] Education and socio-economic status are important variables that might also interplay with the feasibility of the AIMS home-video method and need to be addressed in further studies.

The dropout rate in this study was considerable which threatens feasibility. However, we investigated both the feasibility of the home video method and the feasibility of applying it longitudinally. We asked parents with a young baby to commit themselves to the study for a period of nine months. All parents who participated in the pilot, delivered one to five adequate home videos, which shows the home-video method itself was feasible for these parents. It was mainly the final questionnaire (T1) which was returned poorly (n = 34). These data indicate that the longitudinal aspect of the study was probably the main reason for dropout. Another limiting factor was that a majority of parents who signed up to participate (filled out the questionnaires and participated in the interviews) were mothers. The young age of some of the participating infants (as low as 1.5 months at the start) might have played a role in this phenomenon. Having maternity leave, Dutch mothers were probably more available and willing to become involved in research than fathers. Although most parents worked together to record the home videos, it was mainly the experiences of the mothers that were collected in both questionnaires and interviews. This is a known limitation in infant studies[34] and it is important to consider because fathers might have different experiences than mothers, especially with regard to digital equipment.

Conclusion

The present study provides evidence that the AIMS home-video method is feasible for participating parents regarding both practical aspects and the understanding of their task. Most identified barriers reported by parents have a practical nature that can be addressed in future applications. The home-video method has the potential to become a valuable E-health addition for both research and PPT practice to monitor infants at risk of developmental motor delay in their own familiar environment.

More research is needed to explore if these findings are applicable to parents with different backgrounds and to parents of infants at risk. How will these parents experience a more explicit role in the assessment of their child's risk for a delay in motor development? Will the active involvement of parents indeed lead to increased awareness and knowledge of motor development? In short, can the AIMS home-video method become more than just a means and become a tool to empower parents who have an infant at risk of developmental delay?

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Author contributions

Marike Boonzaaijer: Conceptualization, Methodology, Investigation, Resources, Data Curation, Formal Analysis, Writing – Original Draft, Project administration. *Floryt van Wesel:* Methodology, Formal Analysis, Writing - Review & Editing. *Jacqueline Nuysink:* Conceptualization, Methodology, Writing - Review & Editing. *Chiel Volman:* Methodology, Formal Analysis, Writing - Review & Editing. *Marian Jongmans:* Conceptualization, Methodology, Formal Analysis, Writing - Review & Editing, Supervision.

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Additional File 3.1. Digital questionnaires (T0 and T1)

Questionnaire for parents (T0): Expectations on the home-video method

Personal data

1. Please enter your child's research code.
2. What is the date of birth of your child?
3. What was the duration of pregnancy?
4. What was the birthweight of your child?
5. What is the birth rank of your child?
6. What is the name of the general practitioner?
7. What is the place of residence of the general practitioner?
8. What is your age?
9. What is the age of your partner?
10. What is your level of education?
11. What is your partner's level of education?
12. What is your relationship with the child (father/mother/other)
13. Because of my work/education, my knowledge of infant motor development is more than average
    - I strongly disagree
    - I disagree
    - Neutral
    - I agree
    - I totally agree

Questions on expectations

14. How much effort do you expect this study will cost you as a parent?
    Give a number from 0–10 (0 = no effort at all, 10 = a lot of effort).

15. How much effort do you expect this study will cost your baby?
    Give a number from 0–10 (0 = no effort at all, 10 = a lot of effort).

16. What is your motivation to participate in this study?
    - I think the study is useful and interesting.
    - I think participation in research project is important.
    - It is nice to know about the motor development of my child.
    - I have a question on the motor development of my child.
    - Other:

17. I expect that the technical aspects of recording the home video (light, distance, camera position) will be easy to carry out.
    - I strongly disagree
    - I disagree
    - Neutral
    - I agree
    - I totally agree

18. I expect that deciding which positions and movements of my child I have to capture on home video will be easy to do.
    - I strongly disagree
    - I disagree
    - Neutral
    - I agree
    - I totally agree

19. I expect that prompting my child to show specific movements will be easy to do.
    - I strongly disagree
    - I disagree
    - Neutral
    - I agree
    - I totally agree

20. Uploading the home videos using the web portal on the computer will not be a problem for me.
    - I strongly disagree
    - I disagree
    - Neutral
    - I agree
    - I totally agree

21. Can you describe your child's current motor development?
    - Faster than average
    - Average
    - Slower than average
    - I have no idea

Questionnaire for parents (T1): Experiences of the home-video method

I Questions on the experiences

1. Please enter your child's research code.

2a. How would you rate the effort of this study for you as a parent?
Give a number from 0–10 (0 = no effort at all, 10 = a lot of effort).

2b. Please explain the given number.

3a. How would you rate the effort of this study for your child?
Give a number from 0–10 (0 = no effort at all, 10 = a lot of effort).

3b. Please explain the given number.

4. It was easy to find an appropriate moment to record the home videos.
    - I strongly disagree
    - I disagree
    - Neutral
    - I agree
    - I totally agree

5. Two weeks' time is enough to find an appropriate moment to record the home videos.
    - I strongly disagree
    - I disagree
    - Neutral
    - I agree
    - I totally agree

6. The instruction videos were clear and understandable.
   - I strongly disagree
   - I disagree
   - Neutral
   - I agree
   - I totally agree

7. The checklists were clear and understandable.
   - I strongly disagree
   - I disagree
   - Neutral
   - I agree
   - I totally agree

8. What device did you use to record the home videos?
   - iPhone smartphone
   - Android smartphone
   - iPad tablet
   - Android tablet
   - Digital camera
   - Other:

9. Uploading the home videos to the web portal from my computer was easy to do.
   - I strongly disagree
   - I disagree
   - Neutral
   - I agree
   - I totally agree

10. Did you experience any difficulties using the device while recording and/or uploading?
    - No
    - Yes, being …

11. To upload the home videos I used:
    - The web portal
    - WeTransfer
    - Other:

12. The technical aspects of the recording were easy to carry out (light, distance, camera position).
    - I strongly disagree
    - I disagree
    - Neutral
    - I agree
    - I totally agree

13. It was clear for me what positions and movements of my child I had to capture on camera.
    - I strongly disagree
    - I disagree
    - Neutral
    - I agree
    - I totally agree

14. Prompting my child to show specific movements was easy to do.
    - I strongly disagree
    - I disagree
    - Neutral
    - I agree
    - I totally agree

15. The feedback I received by email was informative enough for me.
    - I strongly disagree
    - I disagree
    - Neutral
    - I agree
    - I totally agree

16. The feedback I received on my baby's motor development gave cause for concern.
    - I strongly disagree
    - I disagree
    - Neutral
    - I agree
    - I totally agree

The following questions only apply to the last time you recorded a home video of your child.

17. How was your baby's behavioral state during the recording?
    - Sleepy, not active
    - Quiet, less active than normal
    - Cheerful, active
    - Irritable, grumpy
    - Upset, crying
    - Other:

18. On the home video my child showed optimal motor behavior.
    - Yes, my child showed his/her optimal motor behavior
    - Yes, and my child even showed new items
    - No, my child did not show optimal motor behavior (please answer question 19)

19. Please indicate what factors you think had a negative influence on your child's motor behavior during the video recording:
    - My child was disrupted by the camera
    - My child was disrupted by noises or bustle
    - My child had some physical inconveniences
    - My child was not in the mood/ right state
    - The presence of other people disrupted my child
    - It was coincidental that my child didn't show optimal motor behavior
    - Other:

20. What do you think about your child's current motor development?
    - Faster than average
    - Average
    - Slower than average
    - I have no idea

21. Who has most care tasks at the moment?
    - Mother
    - Father/partner
    - Mother and father/partner have equal caring tasks
    - Other:

22. Can you indicate your baby's residency for the days of an average week?
    (at home, with grandparents, daycare, other)

23. Can you indicate factors that might have influenced your infant's motor development during this study? Please indicate at what age.
    (Illness, treatment by a (para)medic, change in family composition, change in living environment, move to a new house, change in daycare)

24. This questionnaire was completed by:
    - Father/partner
    - Mother
    - Other:

Additional File 3.2. Semi-structured topic list interview

Introduction about the interview

I The instruction

- Beforehand, was it clear to you what was expected?
- How much time did you need for preparation?
- What did you think about the instruction videos?
- What did you think about the checklists?

II Recording and uploading the home video

- Please describe how the recording of the home video came about?
- Which digital device did you use to make the recording?
- What did you think about the instructions: were they clear?
- What were your experiences uploading the recordings?

III Handling and prompting the infant

- How did you feel about handling your baby and prompting movements according to the instructions?
- Were you able to find an appropriate time for recording?
- Was it clear to you which postures and movements you were supposed to record?
- Was your baby able to show his/her optimal motor performance during the recording?

IV Feedback on motor development

- What do you think about the feedback you received on the motor development of your baby?
- Did the feedback influence your actions or thoughts towards/about your baby?

V Experiences in general

- How did you experience your participation in this research project in general?
- How was it to make multiple recordings over a time of 9 months?
- What motivated you to stay involved?
- How could we improve parental compliance even more?

- What are your thoughts on the safety of the video recordings of your child and privacy issues?
- Do you think the home-video recording method is feasible for all parents?
- What do you think about the home-video method to assess an infant's motor development?
- Do you have ideas to improve the home-video method?
- Is there anything you would like to add or comment on?

Additional File 3.3. Checklist I–III AIMS home-video method

Checklist I The baby is not rolling over yet

This checklist can be used during filming. Don't forget to watch the instruction video.
In this checklist you will find:
- The movements and positions we want you to capture on the home video.
- Tips to pay attention to, so your home video can be used to assess motor performance.

Check	Tips
General	☐ We will assess the motor skills of your baby, so let him/her move freely and try not to help with your hands.
	☐ A good way to start the video is to film spontaneous movements of your baby; please don't elicit movements with toys or sounds right away.
	☐ During filming, make contact with your baby like you always do.
	☐ The positions we ask you to film do not have to be filmed in the order displayed. Breaks can be taken if that's desirable.
	☐ If you make the home video with your smartphone, the phone has to be in a horizontal position.
	☐ During filming, your baby should only be wearing a body suit.
Environment	☐ Try to film with the light source behind you.
	☐ Please film your baby on the floor and make sure the under layer is firm and prevents sliding.
Duration and timing	☐ Please make sure you have 10-15 minutes on tape. The maximum length of the home video is 30 minutes.
	☐ Try not to make multiple short video shots. We prefer longer shots.
	☐ When your baby is getting tired or discomforted, it is better to stop and try filming again another time.

* The development of this checklist was part of a grant research project (2013-53p).

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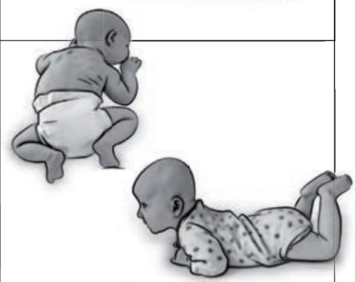
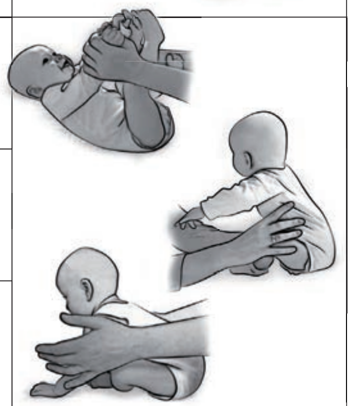
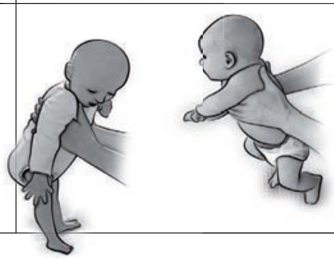
Checklist I The baby is not rolling over yet

Position	Check the tips	Camera position from the	
Supine	☐ Film a few minutes in supine position without a toy.	side bottom	
	☐ Present a little toy above your baby, to elicit reaching and/or grabbing.	side bottom	
Prone	☐ Lay your baby down in prone position with his/her hands at shoulder level. Film a few seconds without making contact. After that make contact with your baby to see if he/she is able to actively raise the head.	side top	
	☐ Present a small toy right in front of your baby.	side top	
Pull to sit	☐ Make eye contact with your baby in supine position, so he/she turns the head to the midline. Then hold the wrists of your baby and pull gently. When the head still lags behind, lay down your baby gently. Repeat this one more time.	side	
Sitting with support	☐ Keep your baby supported in the sitting position and see if you can make eye contact.	front side	
	☐ See if your baby can sit without support for a brief moment. Your baby may use the arms as support forward. Keep your hands close by, sitting is not a stable position yet.	front side	
Supported standing	☐ Hold your baby between the pelvic and the shoulders. Let the feet touch the floor and see if your baby takes some weight on the feet or toes.	front side	

Checklist II The baby is rolling over and starting to move

This checklist can be used during filming. Don't forget to watch the instruction video.
In this checklist you will find:
- The movements and positions we want you to capture on the home video.
- Tips to pay attention to, so your home video can be used to assess motor performance.

Check	Tips	
General	☐ We will assess the motor skills of your baby, so let him/her move freely and try not to help with your hands.	
	☐ A good way to start the video is to film spontaneous movements of your baby; please don't elicit movements with toys or sounds right away.	
	☐ During filming, make contact with your baby like you always do.	
	☐ The positions we ask you to film do not have to be filmed in the order displayed. Breaks can be taken if that's desirable.	
	☐ If you make the home video with your smartphone, the phone has to be in a horizontal position.	
	☐ During filming, your baby should only be wearing a body suit.	
Environment	☐ Try to film with the light source behind you.	
	☐ Please film your baby on the floor and make sure the under layer is firm and prevents sliding.	
Duration and timing	☐ Please make sure you have 10-15 minutes on tape. The maximum length of the home video is 30 minutes.	
	☐ Try not to make multiple short video shots. We prefer longer shots.	
	☐ When your baby is getting tired or discomforted, it is better to stop and try filming again another time.	

* The development of this checklist was part of a grant research project (2013-53p).

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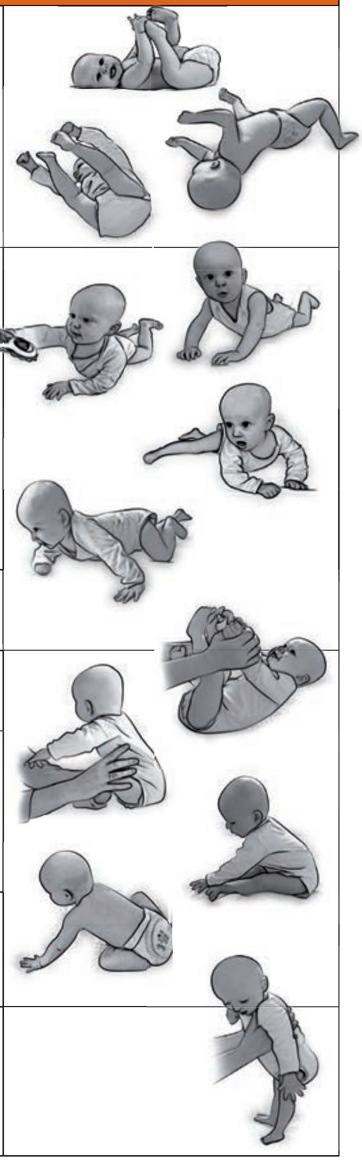
Checklist II The baby is rolling over and starting to move

Position	Check the tips	Camera position from the	
Supine	☐ Please film a few minutes in supine position without a toy	side bottom	
	☐ Present a little toy above your baby, in that way you can elicit reaching and/or grabbing		
	☐ Present a toy beside the head of your baby, maybe he/she will roll over		
Prone	☐ If needed, help your baby to lay down in prone position. Film the spontaneous movements for a short while.	side top	
	☐ After that, present a toy in the sight of your baby; in front of him/her.		
	☐ Present a toy above the head and shoulders. Try to elicit reaching or grabbing the toy by leaning on one arm. Try this at both sides.		
	☐ Present a toy and move it in a circle around your baby so he/she will follow it. Now your baby is dialling on his/her belly.		
	☐ If you know your baby can move forward on the belly, try to capture this.	side	
Pull to sit	☐ Hold the wrists of your baby and pull gently to the sitting position. Please film this movement one more time.	side	
Supported sitting	☐ Keep your baby supported in sitting position and see if you can make eye contact.	front side	
	☐ See if your baby can sit on his/her own for a brief moment.		
	☐ If you know your baby can transfer from sitting to supine position, film this.	side	
Standing	☐ Hold your baby between the pelvic and the shoulders. Let the feet touch the floor to see if he/she takes some weight on the feet.	front side	

Checklist III The baby is crawling and / or walking

This checklist can be used during filming. Don't forget to watch the instruction video.
In this checklist you will find:
- The movements and positions we want you to capture on the home video.
- Tips to pay attention to, so your home video can be used to assess motor performance.

Check	Tips
General	☐ We will assess the motor skills of your baby, so let him / her move freely and try not to help with your hands.
	☐ A good way to start the video is to film spontaneous movements of your baby; please don't elicit movements with toys or sounds right away.
	☐ During filming, make contact with your baby like you always do.
	☐ The positions we ask you to film do not have to be filmed in the order displayed. Breaks can be taken if that's desirable.
	☐ If you make the home video with your smartphone, the phone has to be in a horizontal position.
	☐ During filming, your baby should only be wearing a body suit.
Environment	☐ Try to film with the light source behind you.
	☐ Please film your baby on the floor and make sure the under layer is firm and prevents sliding.
Duration and timing	☐ Please make sure you have 10-15 minutes on tape. The maximum length of the home video is 30 minutes.
	☐ Try not to make multiple short video shots. We prefer longer shots.
	☐ When your baby is getting tired or discomforted is better to stop and try filming again another tin

* The development of this checklist was part of a grant research project (2013-53p).

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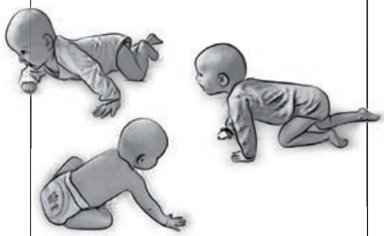
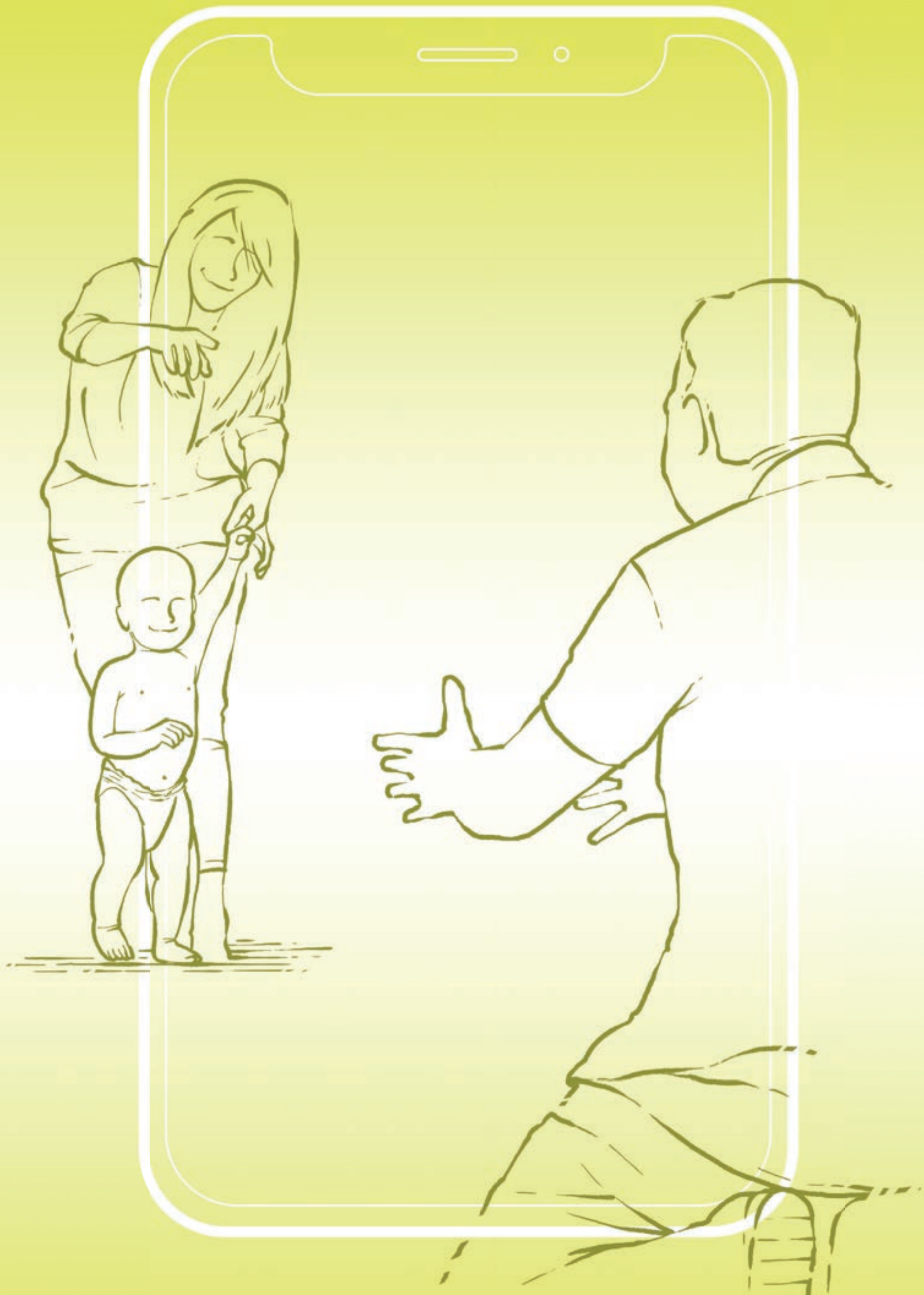
Checklist III The baby is crawling and / or walking

Position	Check the tips	Camera position from the	
Supine	☐ Please, turn on the camera when you take off your baby's clothes in supine position.	side	
	☐ At this age, your baby can roll over very easily. Try to capture this movement to both sides.		
Prone	☐ Capture your baby moving forward, this can be crawling or creeping.	side	
	☐ Try to film the transfer from sit to crawling.		
Sitting	☐ Film your baby while he / she is transferring to sit. Let it play with some toys in this position.	side	
	☐ Present a toy to your baby at both the left and the right side and out of reach, so he / she has to turn to reach for the toy.		
Standing	☐ Put some toys on the couch or the table. If your baby does not pull up to a standing position, help your baby on the feet.	walking around	
	☐ Encourage walking along the couch or table by replacing toys or making contact.		
	☐ Is your baby capable to transfer from a standing position to a sitting position? Try to capture this.		
	☐ If your baby is capable to stand or walk without support, try to capture this.		
	☐ Playing in a squatted position without support is the final position to film.		

PART II



Chapter 4

Modeling a gross motor curve of typically developing Dutch infants from 3.5 to 15.5 months based on the Alberta Infant Motor Scale

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ABSTRACT

Background: Interindividual variability in gross motor development of infants is substantial and challenges the interpretation of motor assessments. Longitudinal research can provide insight into variability in individual gross motor trajectories.

Purpose: To model a gross motor growth curve of healthy term-born infants from 3.5 to 15.5 months with the Alberta Infant Motor Scale (AIMS) and to explore groups of infants with different patterns of development.

Methods: A prospective longitudinal study including six assessments with the AIMS. A Linear Mixed Model analysis (LMM) was applied to model motor growth, controlled for covariates. Cluster analysis was used to explore groups with different pathways. Growth curves for the subgroups were modelled and differences in the covariates between the groups were described and tested.

Results: In total, data of 103 infants was included in the LMM which showed that a cubic function ($F(1,571) = 89.68$, $p < .001$) fitted the data best. None of the covariates remained in the model. Cluster analysis delineated three clinically relevant groups: 1) Early developers (32%), 2) Gradual developers (46%), and 3) Late bloomers (22%). Significant differences in covariates between the groups were found for birth order, maternal education and maternal employment.

Conclusion: The current study contributes to knowledge about gross motor trajectories of healthy term born infants. Cluster analysis identified three groups with different gross motor trajectories. The motor growth curve provides a starting point for future research on motor trajectories of infants at risk and can contribute to accurate screening.

INTRODUCTION

In the first two years of human life, gross motor development is the most important indicator of wellbeing and general development[1] and therefore of great importance for early developmental screening. While former theories like Gesell[2] and McGraw[3] assumed uniformity in terms that all infants achieve motor milestones in more or less the same sequence and pace, it has become increasingly clear that variability between and within infants are typical features of motor development in infancy.[4,5]

Since the '90s, the dynamic systems theory (DST) provides a foundation for explaining variability in motor development by stating that continuous changes in an infant's body and environmental changes provide different opportunities for development.[5] In this light, numerous studies investigated the impact of child and environmental factors on gross motor development, such as birth weight and gestational age,[6] birth order,[7,8] caregiving practices,[9] affordances in the home,[10] maternal age and education,[11] and the influence of parents' mental wellbeing and beliefs.[11,12] The result of this complex interplay of genetic and environmental factors is that the gross motor development of a child is non-linear in nature. Therefore, to reliably chart motor development and capture the true shape of development, multiple time points have to be assessed and important factors known to be associated with gross motor development should be included in the analysis.[5,13]

Few longitudinal studies have been conducted to investigate intra-individual variability in gross motor trajectories. Darrah and colleagues[14,15] found that babies whose gross motor development was assessed from birth to independent gait showed great variability in their percentile rankings on the Alberta Infant Motor Scale (AIMS). In a study on 83 children from 9 months till 5.5 years they reported that the percentile rankings of the Peabody Developmental Motor Scales (PDMS) were best represented by non-linear equations, even though the within-subjects variability decreased after infancy.[15] In a longitudinal study on term (n = 30) and preterm born infants (n = 59), motor performance and movement quality were assessed five times with the Infant Motor Profile (IMP) from 3 to 18 months. Heineman and colleagues[16] found higher IMP scores and a smaller within-participant variability in the term group in comparison to the preterm group. A quadratic function of age was found to be the best fit for the data of the total group in a mixed-effects model.

In addition to intra-individual variability in gross motor development, variability between infants has also been observed. For example, the World Health Organization demonstrated a large spread in the time of motor milestone attainment in 816 infants from five countries.[17] The ages at which infants achieved the milestone 'sitting without support' varied from 3.8 months to 9.2 months. The age at which infants started to walk independently showed a range of more than 9 months, from 8.2 months to 17.6 months.

Other studies have tried to identify variation in different pathways in infant motor development. Eldred et al.[18] reported four clusters of infants with similar trajectories of percentile rankings on the PDMS within a group of 66 infants aged 9 months to 5.5 years. The scoring patterns of percentile rankings over time were described as 'robust scores', 'decreasing scores', 'increasing scores', and 'low scores'. Another study that applied latent class analysis on a cohort of 1254 infants, revealed a model with three classes of infants with similar gross motor pathways on the age-equivalent-normcores of the Ages and Stages Questionnaire (ASQ-II) from 4 to 24 months namely, the 1) 'high stable class', (80% of the infants), 2) the 'U-shaped class', and 3) the 'late bloomers'.[19] Nishimura and colleagues[20] used latent class growth analysis in a birth cohort study (*N* = 952) and found five distinct trajectory patterns in the gross motor scale of the Mullens Scales of Early Learning (MSEL) on seven assessments between 1 and 24 months. The five classes were described as high normal, normal, low normal, delayed, and markedly delayed.

In summary, these longitudinal studies suggest that both intra-individual and inter-individual variability in gross motor trajectories are indeed characteristics of typically developing infants. Clinically, interpreting variability is a challenge for pediatric physical therapists (PPT).[18] When motor development does not follow a stable pattern over time, early prediction of later development would not be reliable. Subsequently, this raises questions about the timing and frequency of developmental surveillance and early intervention.[21]

Gross motor trajectories of healthy term-born infants have not yet been studied in the Netherlands. In previous Canadian research conducted by Darrah and colleagues,[14] culturally specific percentile scores of the AIMS were used to examine intra-individual variability. Converting motor outcomes of Dutch infants into percentile scores based on cross-sectional Canadian norms seems not appropriate in the light of cross-cultural differences.[22,23] Besides, methodological research advocates the use of change scores to describe growth in motor outcome over time, rather than derived percentile scores intended to provide a normative evaluation of skills.[24]

So, in contrast to previous research, this study aims to model a motor growth curve using the raw test scores of the AIMS in typically developing (Dutch) infants. This method is expected to shed new light on motor development by showing growth beyond the norm scores. The following control variables will be included in the analysis: birth weight, gestational age, birth order, and maternal education, and maternal employment.

The growth curve can serve as a point of departure for future research on developmental trajectories of Dutch infants at risk for delays such as preterm born infants or infants with congenital heart diseases.[25,26] Furthermore, identifying different pathways of typically developing infants can support clinicians to estimate whether or not the observed motor behavior is within the normal range.

Therefore, the specific objectives of this study were:

- To model motor growth in a population of typically developing Dutch infants from 3.5 to 15.5 months using AIMS raw scores.
- To explore different patterns in gross motor trajectories within a population of typically developing infants from 3.5 to 15.5 months.

PARTICIPANTS AND METHODS

Design and participants

This study had a prospective longitudinal design. Parents of healthy term-born infants were invited through open registration from May 2016 and April 2018 leading to a convenience sample. Recruitment took place by distributing flyers at birth centers, day-care centers, well-baby clinics, and maternity care offices in the larger cities of the Netherlands. Also, communication channels on social media were used to inform parents about the study. Infants born before 37 weeks of gestational age or diagnosed with pathology were not eligible to enter the study. Only parents with sufficient understanding of the Dutch language to read the informed consent and the instructions were included in the study. Either parents or legal representatives had to sign informed consent. Ethical approval for the study was obtained from the medical ethics committee of the University Medical Centre in Utrecht, the Netherlands (METC number 16/366C).

Procedure and measures

To collect data on parent and infant characteristics, online questionnaires were sent by email at the infants' ages of 3.5, 5.5, 15.5, and 18 months. Infant characteristics included gender, birth weight, gestational age, and birth order (1 = firstborn, 2 = second-born, 3 = third and fourth child). Furthermore, questions about perinatal events and treatment by a paramedical or medical specialist were also collected by parental reports.

Information about parents that was obtained included age, education, occupation, and native language. Parental age was reported in five categories: 20–24, 25–29, 30–34, 35–39, and 40–45 years. The highest level of parental education was reported in five categories: no education, primary, lower secondary, higher secondary, and tertiary education which is equivalent to a university degree. The occupation of parents was categorized according to a Standardized Classification of Professions in the Netherlands (SCB, 2010) into six categories (Table 4.1).

Gross motor development was assessed with the AIMS at 3.5, 5.5, 7.5, 9.5, 12.5, and 15.5 months. Internationally, the AIMS has been a preferred measure for over 30 years[25] and is considered reliable and valid with Intraclass Correlation Coefficients (ICC) of 0.992 and 0.987 for inter-and intra-rater reliability, respectively.[28] In terms of concurrent validity, ICCs were established of 0.98 with the Bayley Scales of Infant and Toddler Development,[29] and 0.97 with the PDMS.[28,30] The AIMS consists of 58 items, divided into 4 subscales: prone (21 items), supine (9 items), sitting (12 items), and standing (16 items). To determine a total raw score, the infants' spontaneous movements have to be observed in the four positions. The total score can be converted to a percentile rank and a *Z*-score. The norm population on which the references are based comprised of 2022 infants from Alberta, Canada observed in 1994.[31] From a Canadian re-evaluation in 2014, the authors concluded that the norm references are still valid for the Canadian population.[32] Lately, in several countries, norm reference studies have been carried out to evaluate cultural validity.[22,33,35] Very recently, Dutch AIMS norm scores were reported based on video observations of 1697 infants.[23]

To enable the collection of longitudinal data on motor development, the AIMS home-video method for parents was used.[36] Parents received instructions (Additional File 3.3) on how to position their baby and what movements to prompt. Parents were notified by email when they had to make a home video and upload it to a secured web portal. Parents were given a two-week window to make the video and reminders were sent once within the window. From the web portal, the videos were assessed with the AIMS by a trained PPT/researcher and parents received feedback on the development of their infant either by email or telephone. Four PPT/researchers, who performed the assessments, attended two training sessions of three hours to ensure the reliability of scoring the AIMS assessments. The agreement between the two main observers was found to be 97.8% on the scored items of eight infants. Adjusting this outcome for chance with Cohen's

Kappa, the agreement was .95, which is almost perfect.[34] During the study, difficulties in scoring were reviewed and discussed to ensure continuing consensus on item level.

The concurrent validity of the AIMS home-video method was established with a mean difference of 0.46 items between live- and video-observations and an excellent ICC agreement of 0.99. The Standard Error of the Measurement was calculated to be 1.48 items and the smallest detectable change was 3.88 items.[36] Parents' experiences with the longitudinal use of the home-video method were evaluated and found to be both feasible and acceptable.[37]

Statistical analysis

First, data were explored visually and descriptive statistics were applied. Initially, Latent Growth Modeling (LGM) was used to model motor growth. However, these models did not adequately fit the data (Appendix 4.1). A Linear Mixed Model analysis (LMM) was considered to be a good alternative because it considers the dependence of repeated measures within one infant and allows for a variable number of observations.[39] When modeling growth in a multilevel model, both variability within and between subjects is taken into account. To determine the overall shape of developmental change, linear, quadratic, and cubic functions were fit according to the strategy suggested by Singer and Willet.[39] The intercept and slope were allowed to vary across individuals. To select the best model, the Likelihood Ratio Test was used.[40] The most parsimonious model was controlled for the infant factors: birth weight, gestational age, and birth order as well as for the maternal factors: age, education, and employment status. To do so, a backward selection of variables was used with a $p < .05$ as selection criteria to control for their effect on the shape of the curve. For these variables, fixed effects were assumed.[38]

After visual inspection of the individual motor trajectories, a hierarchical cluster analysis was applied to identify different groups of infants showing similar patterns in gross motor development, based on the AIMS raw motor scores at 3.5, 5.5, 7.5, 9.5, 12.5, and 15.5 months. To group infants with similar trajectories, the between-groups linkage method was applied with a Euclidean distance measure.[41] The optimal number of clusters was determined by a dendrogram and an agglomeration schedule. Subsequently, a K-means cluster analysis was computed to fine-tune the clusters. The characteristics of the groups were described and one-way ANOVAs and Kruskal-Wallis tests with post hoc analysis were applied to indicate differences in continuous and categorical variables between the groups. Finally, LMM was applied to model growth curves of the developmental clusters including their interaction with time. Statistical analyses were performed with IBM SPSS Statistics for Windows, Version 25.0.

RESULTS

In total, trajectories of 103 infants were included in the analysis. Of these, 18 infants missed one assessment, ten infants missed two assessments and two infants missed three assessments of the six assessments in total. Since the primary reason for the missing assessments concerned holidays, moving to a new house, or the busyness of parents, the missing data were considered random. Over time, there was a slight increase in missing data, which is common in longitudinal studies. The maximal attrition rate of 14.6% at 15.5 months is within acceptable limits.[42] Because LMM allows for the inclusion of subjects with missing data,[38] only infants with < 3 assessments available (*n* = 12) were excluded because fitting a higher-order function would not be possible on only two time points. The characteristics of infants and parents are displayed in Table 4.1.

Individual trajectories of gross motor development

The mean total raw scores on the AIMS are displayed in Supplementary Table S4.1 and the individual trajectories in Figure 4.1. Unidirectional growth is visible for all infants in a sigmoid-shaped curve and accelerations and decelerations at different times in the

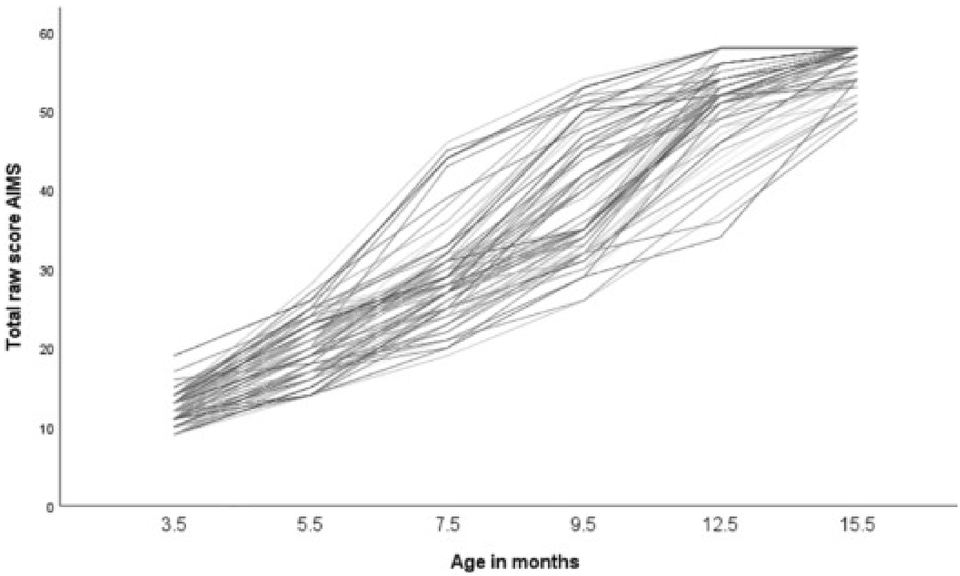

Figure 4.1 Individual trajectories 3.5–15.5 months in raw AIMS scores [0–58] (*N* = 103).

individual growth curves. At 3.5 months, the standard deviation of the raw AIMS scores is lower compared to the assessments that follow. A ceiling effect of the test is present at 15.5 months because 51 of 88 infants had reached the total score of 58 items. At 12.5 months, 10 of 91 infants had reached the total test score. Between 7.5 and 9.5 months the mean change score was the largest, amounting to 11.2 items (5.6 items/month), indicating that most infants accelerate in their motor growth between these time points.

Table 4.1 Demographic characteristics of infants and parents

Infant characteristics		*M* (*SD*)	Range	*N*
Gender	Female Male	64 (61.5%) 39 (38.5%)		103
Birth weight		3528.3 grams (409.3)	[2780–4560 g]	103
Gestational age		39.9 weeks (1.3)	[37–42 weeks]	103
Birth order	1st 2nd 3rd 4th	55 (52.9%) 38 (36.5%) 8 (8.7%) 2 (1.9%)		103
Perinatal events according to parents[1]	No Yes	85 (82.5%) 18 (17.5%)		103
Parent characteristics		**Maternal**	**Paternal**	***N***
Parental age	20–24 years 25–29 years 30–34 years 35–39 years 40–45 years Single parent (NA)	2 (1.9%) 16 (15.5%) 53 (51.5%) 24 (23.3%) 8 (7.8%)	1 (1%) 13 (12.6%) 31 (30.1%) 42 (40.8%) 12 (11.7%) 2 (1.9%)	103
Parental education	No education Primary Secondary lower Secondary higher Tertiary Single parent (NA)	0 (0%) 0 (0%) 1 (1%) 13 (12.6%) 89 (86.4%)	2 (1.9%) 0 (0%) 1 (1%) 15 (14.4%) 83 (79.7%) 2 (1.9%)	103
Parental professional classification	No profession Elementary Lower Secondary Higher Scientific	6 (5.8%) 1 (1.0%) 4 (3.9%) 15 (14.6%) 54 (52.4%) 23 (22.3%)	2 (1.9%) 1 (1.0%) 4 (3.9%) 13 (12.6%) 62 (60.2%) 21 (20.4%)	103
Native language	Dutch Other	99 (96.1%) 4 (3.9%)		103

[1] Perinatal events reported by parents were: delivery by vacuum pump, maternal blood loss during delivery, non-progressing birth, emergency Cesarean section, uterus rupture and releasing placenta, maternal high blood pressure, meconium in amniotic fluid.

4

If the total AIMS scores of this sample are compared to both the Canadian norms[28] and the recently introduced Dutch AIMS norms[23] on a *p*5 cut-off point, it appears that none of the participating infants scored below the 5th percentile at any time point on the new Dutch AIMS norms. If we look at the Canadian norms, a different picture emerges. At each time point, except for 3.5 months, there is a considerable number of infants scoring below the *p*5: 11% at 5.5 months, 6% at 7.5 months 24% at 9.5 months, 20% at 12.5 months, and 42% at 15.5 months.

Modeling a gross motor growth curve of infants aged 3.5 to 15.5 months

A nonlinear function, a cubic polynomial, yielded the best fit for the overall data ($F(1,571) = 89.68$, $p < .001$). The curve represents the average scores predicted by the model and is characterized by an initial slow growth in AIMS scores followed by an overall acceleration till 12.5 months with a subsequent deceleration from 12.5 months to 15.5 months (see Figure 4.2). Using a backward selection ($p < .05$) the covariates were added to the model. None of the covariates remained in the final model even though the overall effect of birth order showed a trend ($F(2,104.83) = 2.35$, $p = .10$) with a marginally significant difference between firstborn infants and infants that are third or fourth in birth order (β = 2.33, *SD* = 1.28, $p = .07$).

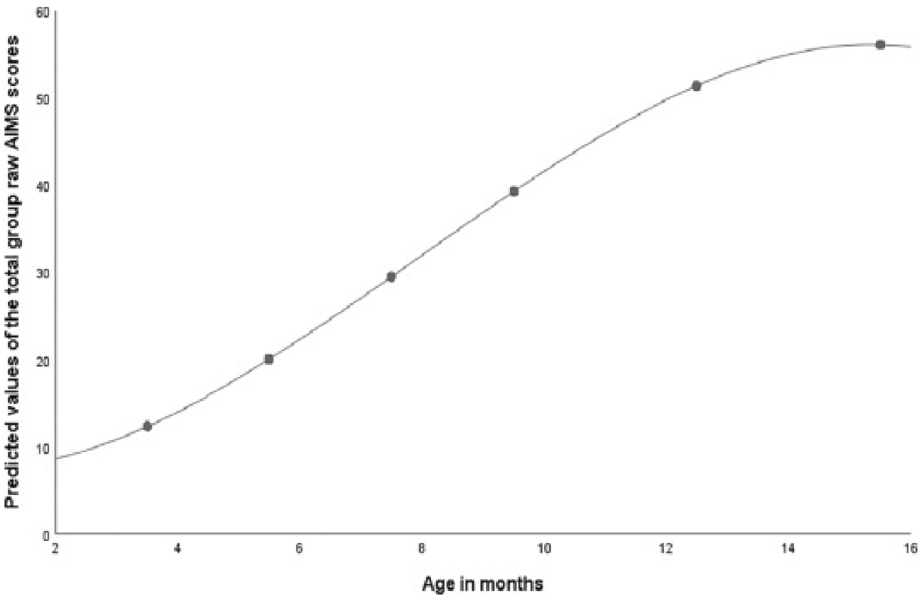

Figure 4.2 Growth curve of gross motor development 3.5–15.5 months (*N* = 103).

Patterns of gross motor trajectories

Cluster analysis delineated three groups, and visual inspection confirmed that these clusters were clinically relevant. The K-means cluster analysis needed seven iterations to converge. The three clusters were labeled as follows:

1. Late bloomers (*n* = 23) (22.3%) who mostly do not start accelerating in motor growth before 9.5 months and although a lot of catching up growth can be observed in this group, about 90% of the infants did not achieve all items on the test at 15.5 months.

2. Gradual developers (*n* = 47) (45.6%), with a more even motor growth. Most children in this group achieve all items before 15.5 months.

3. Early developers (*n* = 33) (32%), who show rapid motor growth before the age of 9.5 months and have achieved all items of the AIMS well before 15.5 months and in some cases before 12.5 months.

Modeling growth curves on developmental groups

The individual growth curves of the Late bloomers (*n* = 23), Gradual developers (*n* = 47), and Early developers (*n* = 33) showed a significant effect of time when the clusters were added to the baseline model (Figure 4.3). Significant interactions between time

4

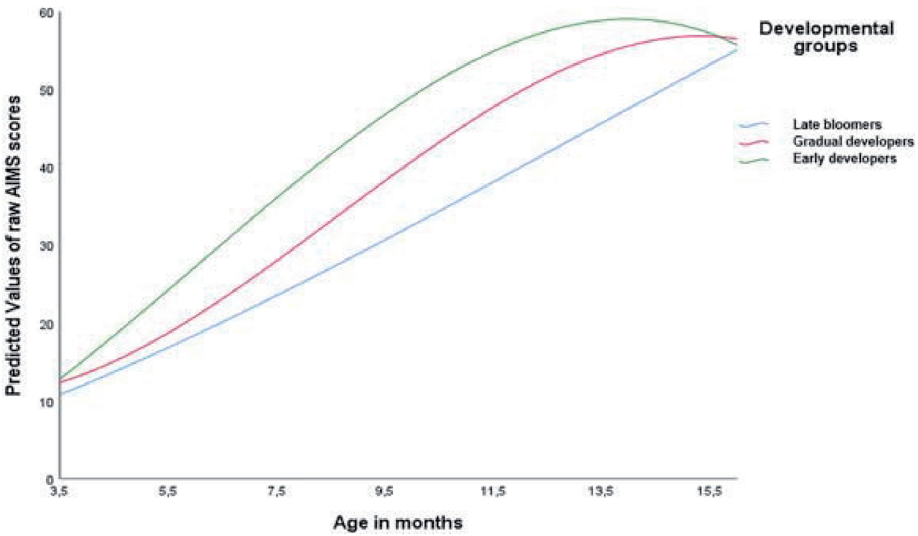

Figure 4.3 Growth curve of gross motor development of Late bloomers, Gradual developers and Early developers 3.5–15.5 months (*N* = 103).

and groups showed that each group follows a unique line and that the slopes are not parallel. The Early developers have higher change scores at the beginning of the curve. At the end of the curve, their change scores diminish due to the ceiling effect of the test. The Late bloomers' change scores are smaller at the beginning with an evident increase towards the end of the curve. The Gradual developers progress in an almost linear manner.

A significant interaction was present between the Early developers and Late bloomers at the end of the curve. The individual variance within the groups was found to be 2.78 items on the 58-item scale of the AIMS (√residuals). The estimates of the cubic growth over time of the groups as well as the total group are provided in Table 4.2.

Table 4.2 Growth curve outcomes for three developmental groups

	Estimate	*SE*	*p*	95% CI	
Intercept	12.78	.57	.000*	11.66	13.89
Time	5.37	.41	.000*	4.57	6.17
$Time^2$	0.24	.08	.005*	0.07	0.40
$Time^3$	-0.03	.00	.000*	-0.04	-0.02
Groups					
Late	-2.04	.85	.016*	-3.70	-0.37
Gradual	-0.45	.71	.527	-1.85	0.95
Groups X Time					
Late	-2.52	.62	.000*	-3.74	-1.30
Gradual	-3.22	.52	.000*	-4.23	-2.20
Groups X $Time^2$					
Late	-0.14	.13	.284	-0.40	0.12
Gradual	0.35	.11	.001*	0.14	0.57
Groups X $Time^3$					
Late	0.03	.01	.000*	0.01	0.04
Gradual	-0.01	.00	.232	-0.02	0.01
Residuals	7.47	.49	.000*	6.56	8.50
Intercept variance	0.43	.44	.323	0.06	3.16

Abbreviations: SE, Standard Error; p, significance; CI, Confidential interval. * $p < .05$.
'Early developers' is the reference group.

The differences in birth weight, gestational age, birth order, maternal education, and maternal employment in the three developmental groups are shown in Table 4.3. Significant differences between the groups were found for birth order, maternal education, and maternal employment. Late bloomers showed a significantly higher mean birth order compared to Gradual developers. Mothers of Early developers had significantly lower education than mothers of Gradual developers. The maternal employment classification of the Gradual developers was significantly higher compared to the Late bloomers (Table 4.3). The mean scores on the AIMS assessments significantly differed between the groups at each time point. This was also the case for the motor milestones achievement of sitting without support, crawling, and independent walking (see Supplementary Table S4.1).

Table 4.3 Differences in gender, birth weight, gestational age, birth order, maternal education and employment in the three developmental groups

	Total group		Developmental groups and comparisons between groups			
	Mean (*SD*) or Number (%)	*N*	Late bloomers *N* = 23 Mean (*SD*)	Gradual developers *N* = 47 Mean (*SD*)	Early developers *N* = 33 Mean (*SD*)	Overall *p*-value
Gender (% female)	61.5%	103	62.5%	66%	54.4%	.653
BW (grams)	3528.28 (409.28)	103	3558.17 (404.96)	3508.68 (389.01)	3534.46 (444.14)	.886
GA (weeks)	39.86 (1.25)	103	39.92 (1.06)	39.77 (1.34)	39.97 (1.26)	.755
Birth order (1–4)	1.58 (.68)	103	1.91[a] (.67)	1.47[a] (.69)	1.50 (.62)	.016*
Maternal age (1–5)	3.14 (.83)	103	3.22 (.90)	3.19 (.80)	3.03 (.83)	.576
Maternal education (1–5)	3.85 (.38)	103	3.82 (.40)	3.98[a] (.15)	3.71[a] (.52)	.005*
Maternal employment (0–5)	3.74 (1.22)	103	3.35[a] (1.30)	3.98[a] (1.19)	3.67 (1.19)	.027*

* Significant differences in post hoc analysis ($p < .05$).
[a] Significant difference between groups with the same letter.

DISCUSSION

This study aimed to model a motor growth curve of healthy term-born Dutch infants from 3.5 to 15.5 months on the AIMS and to examine patterns in inter-individual motor growth. The trajectories showed unidirectional growth in motor scores with individually-timed accelerations and decelerations. A growth curve with a cubic function was the best fit for the longitudinal data of 103 infants. No significant effects were found for the control variables. Three groups with distinct gross motor patterns were identified:

1) Early developers, 2) Gradual developers, and 3) Late bloomers. Significant interaction with time was found. Testing background variables between the groups, significant differences for maternal education, maternal employment, and birth order were found.

To model a gross motor curve

The main objective of this study was to model a gross motor curve for Dutch infants based on the AIMS. Initially, LGM was applied because this technique includes intra-individual correlation and can analyze the effect of a predictor variable on the developmental trajectory of individuals. LGM revealed that neither linear, cubic nor quadratic, or sigmoidal growth functions adequately fitted the data (see Appendix 4.1) probably because of the small sample size and a lack of variance at the beginning and the end of the trajectories. Even though LMM estimates covariates effects more straightforwardly than LGM does, LMM proved to be a good alternative because of the possibility to include cases with missing data.[43]

In a study by Rosenbaum and colleagues,[44] motor growth curves were created for children with Cerebral Palsy (CP) based on the Gross Motor Function Measurement using LMM. Five distinct curves were found, based on 2632 assessments of 657 children with CP. Rosenbaum et al.[44] concluded that the motor growth curves provided means for prognosis and planning interventions for clinicians. In our study, the motor growth curve is based on 571 assessments of 103 infants. Even though the motor growth curve has prognostic value because of the longitudinal nature of the data, a larger sample size including infants at risk would be needed to create a more robust growth curve of gross motor development for Dutch infants including cut-off points.

In the LMM analysis, no main effect of the covariate birth order on motor development was found although a trend was revealed (p = .10), and a post hoc test showed that the difference between the motor development of firstborn infants and infants with a birth rank of > 2 just did not reach significance (p = .07). Testing differences between the three developmental groups, the mean birth order of the Late Bloomers was significantly higher (M = 1.91, SD = .67) in comparison to the mean birth order of the Gradual developers (M = 1.47, SD = .69). Both these findings imply that in a larger sample, the variable birth order may very well show a significant effect on the shape of the gross motor curve.

The finding that infants with a higher birth order were less advanced in their motor development compared to infants that were lower in birth order is in agreement with findings from earlier studies.[7,8,10] According to the prevailing theoretical concept, the explanation for the delay is that with the presence of siblings, parents' resources are more limited.[8] Parents have to divide their time and attention between the siblings, which can result in a less stimulating environment and therefore causing a more delayed motor development for the youngest sibling. In contrast, the competing imitation theory expects a positive influence of the presence of older siblings caused by the enriched environment the older sibling provides in the opportunities of imitation of behavior and play. However, there is no evidence yet that confirms that infants engage in new motor repertoire based on imitation.[8] Berger and Nuzzo[8] found evidence that the impact of older siblings on gross motor development could be both negative and positive, and might depend on unique family characteristics like the age differences between the siblings and the parental expectations regarding motor development. Based on the results of this study and the above-described evidence from previous cross-sectional studies, the role of birth order in gross motor development deserves more attention in future research.

In the present study, motor development was not predicted by any of the child factors or environmental factors. This might be explained by the homogeneous composition of the sample that consisted solely of term-born infants and parents who were generally higher educated. A large body of longitudinal research into the motor development of prematurely born infants, shows that child factors such as low birth weight and a short gestation period have a major and long-term negative impact on motor development.[6,24,26] This in contrast to the impact of environmental factors on motor development, which is not so evident and seems to be more transient.[9,10,45] Perhaps the less pronounced impact of environmental factors only becomes apparent when high-impact factors of the child are absent, as is the case in the present study with only term-born infants. This assumption is in line with the study of Roze and colleagues,[46] who found that the development of healthy term-born children appeared to be more susceptible to variations in environmental factors such as maternal social economic status than factors within the child.

Explore different patterns in gross motor trajectories

The second aim of this research was to explore different patterns in gross motor trajectories. Cluster analysis provided a means to confirm the presence of groups in the sample. Several studies analyzing developmental data of infants also reported the opportunities of this analysis to identify infants at risk that would benefit from early intervention.[18,47]

As in previous studies in which three,[19] four,[18] or five[20] different groups of motor trajectories were identified, three groups were identified in the present study. However, because the measuring instruments and the statistical techniques to identify groups of developmental trajectories are quite different, the findings of the studies are difficult to compare with the findings of the current study. Valla et al.[19] and Nishimura et al.[20] both used Latent Class Analysis in large cohort-based populations on outcomes on the gross motor domains of the ASQ-II and the MSEL, respectively. Despite these differences and the description of five classes, the classes that Nishimura et al.[20] identified in the normal range (high normal, normal, and low normal) are comparable to the three classes that were found in the current study. The cohort-based inclusion of infants at risk explains the presence of the extra two groups in the study of Nishimura et al.[20] that are not present in this study: delayed and markedly delayed.

Even though Valla and colleagues[19] also identified three groups in their sample that included infants at risk, the use of the ASQ-II makes the results hard to compare. The ASQ-II is a parent-completed developmental screening instrument that evaluates gross motor development on six age-specific items. Although the ASQ-II is a useful diagnostic tool to observe developmental delay, it does not assess gross motor behavior in a direct and more specific manner as the AIMS does. Eldred et al.[18] applied a cluster analysis on the percentile scores of the PDMS and reported the identification of four distinct groups. Even though the analysis to identify groups and the population were quite similar to the present study, the use of the percentile scores, especially the increase and decrease of percentile scores, make a comparison of the outcomes difficult.

The identification of the Late bloomers is also relevant for clinical practice. This group, which made up more than 20% of the total sample, is very likely to be seen in practice because of the delayed pathway they follow. From 9.5 to 15.5 months, between 70% and 85% of the Late bloomers scored at least once below -1.65 *SD* on the Canadian AIMS norms.[31] Despite this slower start, most Late bloomers caught up in their motor growth and started walking at a mean age of 16 months.

When we applied the new Dutch AIMS norms, none of the Late bloomers scored below the *p*5. This finding is notable and could be partly explained by the inclusion of about 7% prematurely born infants in the Dutch norm sample while our sample consisted only of healthy term-born infants. Besides this, the new Dutch AIMS norms are considerably lower than the Canadian norms.[23] Despite the very recent introduction of the new Dutch AIMS norms, the Canadian norms values are still much applied in Dutch clinical practice. Therefore, we think it is important to inform PPTs, paediatricians, and parents, that most of these Late bloomers, despite their poorer progress in the first 9.5 months, catch up in motor development and probably do not need intervention.

Future longitudinal research should also include infants at risk for a motor delay to confirm the presence of the three groups of typically developing infants that were identified in this sample. In addition, this longitudinal study showed once more that results from one single assessment should be interpreted with caution and that clinical reasoning should also include the parents' request for help and a qualitative motor observation.[48]

Strengths and limitations

The longitudinal design from 3.5 to 15.5 months with six assessments on gross motor behavior, adds to the strength of this study. As the attrition remained within acceptable limits, this study also confirms that collecting data longitudinally using home videos made by parents is feasible. The AIMS home-video method provided observations of infant motor behavior that were ecologically valid and with a low burden for both infants and parents.[37] The videos of the assessments enabled deliberation on difficult items between researchers, which increased the reliability of the motor assessment scores. Because time-scheduled home visits were not necessary, investment in time and costs were low.

There were also some limitations to the use of the AIMS in this study. Firstly, at the age of 12.5 months, the distribution of the AIMS raw scores was skewed due to the ceiling effect of the test. This skewness increased at 15.5 months when the majority of infants had reached the end of the test which decreased the discriminative value of the outcomes. With most items located roughly between the ages of five to twelve months, it was confirmed that the AIMS is less sensitive at the beginning and the end of the test.[27,28] Subsequently, the shape of the individual trajectories is partly the product of the distribution of items in the four subscales of the AIMS. Therefore, it is important to keep in mind that these motor trajectories are based on the AIMS measurement tool specifically.

Modeling growth curves for Late bloomers, the Gradual developers, and the Early developers was challenging due to the smaller sample size of each group. The significant interaction between Early developers and Late bloomers is more the product of the chosen model, a cubic polynomial, than a reflection of reality. At 15.5 months, the group of Early developers is too small to pull the cubic function into the straight line that represents the ceiling effect of the test. Despite this, the chosen model was the best approximation of reality.

The generalizability of the outcomes beyond the study sample should be carefully considered due to the small size and an overrepresentation of parents with advanced education. Even though the evidence is inconclusive,[49] several studies do report maternal education as a factor that is associated with gross motor development.[11,45,50]

Implications for future research

Further longitudinal research is required with both healthy infants and infants at risk for delay. Digital innovations should be applied to increase the feasibility of data collection to enable researchers to follow large representative samples in and outside Western society. Research questions should be twofold: 1) to contribute to a deeper understanding of factors that are associated with gross motor development in typically developing infants to develop effective interventions, and 2) to model growth curves that are both culturally- and illness-specific to guide professionals in the field.

CONCLUSION

LMM proved to be a useful statistical technique to model gross motor curves of AIMS scores. Applying cluster analysis, three groups with different gross motor trajectories were identified in the data: Early developers, Gradual developers, and Late bloomers. The distinction of these groups within a sample of typically developing infants is clinically relevant because this underlines the presence of variation in gross motor development within the normal range. Furthermore, this study shows that modeling gross motor growth curves is an interesting point of departure for follow-up studies in populations of infants at risk for delay. The development of illness-specific gross motor profiles will improve clinical decision-making for PPTs and pediatricians. It will also support parents to build adequate expectations of their baby’s development.

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Highlights

- A non-linear cubic function best describes the AIMS motor growth curve of infants aged 3.5–15.5 months.
- None of the child- and environmental factors had a significant effect on the motor growth curve.
- Cluster analysis identified three groups: Early developers, Gradual developers and Late bloomers.

Author contributions

Marike Boonzaaijer: Conceptualization, Methodology, Investigation, Resources, Data Curation, Formal Analysis, Writing – Original Draft, Project administration. *Ora Oudgenoeg-Paz:* Conceptualization, Validation, Methodology, Formal Analysis, Writing - Review & Editing. *Imke Suir:* Conceptualization, Investigation, Writing - Review & Editing. *Paul Westers:* Methodology, Formal Analysis. *Jacqueline Nuysink:* Conceptualization, Methodology, Writing - Review & Editing. *Chiel Volman:* Conceptualization, Methodology, Formal Analysis, Writing - Review & Editing. *Marian Jongmans:* Conceptualization, Methodology, Writing - Review & Editing, Supervision.

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Supplementary Table S4.1 AIMS total scores and age of milestone achievement of the total group and developmental groups

	Total group		Developmental groups and comparisons between groups			
	Mean (*SD*) or Number (%)	*N*	Late bloomers *N* = 23 Mean (*SD*)	Gradual developers *N* = 47 Mean (*SD*)	Early developers *N* = 33 Mean (*SD*)	Overall *p*-value
AIMS 3.5 months	12.31 (1.99) [8-19]	95	10.65 (1.53) [8–14]	12.13 (1.59) [8–14]	13.88 (1.56) [12–19]	< .001*
AIMS 5.5 months	19.85 (3.54) [11–28]	103	17.00 (3.12) [11–25]	19.45 (2.69) [14–25]	22.61 (2.78) [17–28]	< .001*
AIMS 7.5 months	29.17 (6.02) [19– 46]	100	23.77 (2.89) [19–29]	27.43 (2.55) [20–32]	35.50 (5.19) [28–46]	< .001*
AIMS 9.5 months	40.29 (8.10) [26–54]	98	30.27 (2.53) [26–36]	38.34 (4.23) [30–47]	49.84 (2.27) [45–54]	< .001*
AIMS 12.5 months	50.87 (5.60) [31–58]	91	42.21 (5.31) [31–50]	51.81 (1.97) [47–58]	55.14 (2.73) [51–58]	< .001*
AIMS 15.5 months	56.36 (2.47) [49–58]	88	53.26 (2.88) [48–58]	56.80 (1.71) [43–58]	57.79 (.77) [54–58]	< .001*
Sitting without support (months)	7.41 (1.43)	63	7.94^{b} (1.67)	7.71^{a} (1.29)	6.53^{ab} (.99)	.004*
Crawling (months)	9.73 (2.08)	63	12.0^{ac} (1.81)	9.67^{ab} (1.30)	7.92^{bc} (1.28)	< .001*
Independent walking (months)	14.11 (2.11)	87	16.04^{ac}(1.60)	14.04^{ab} (1.59)	12.34^{bc} (1.48)	< .001*

Abbreviation: SD, Standard deviation.
* Significant differences in post hoc analysis ($p < .05$). [abc] Significant difference between groups with the same letter.

APPENDIX 4.1 LGM ANALYSIS

In the first step, a model modeling linear growth was constructed. In this model data from all measurements, moments had a loading of 1 on the intercept and a linearly increasing loading on the slope. This model failed to converge as the covariance matrix includes negative values. This problem often arises when the sample size is small or if the model is a very poor fit to the data.

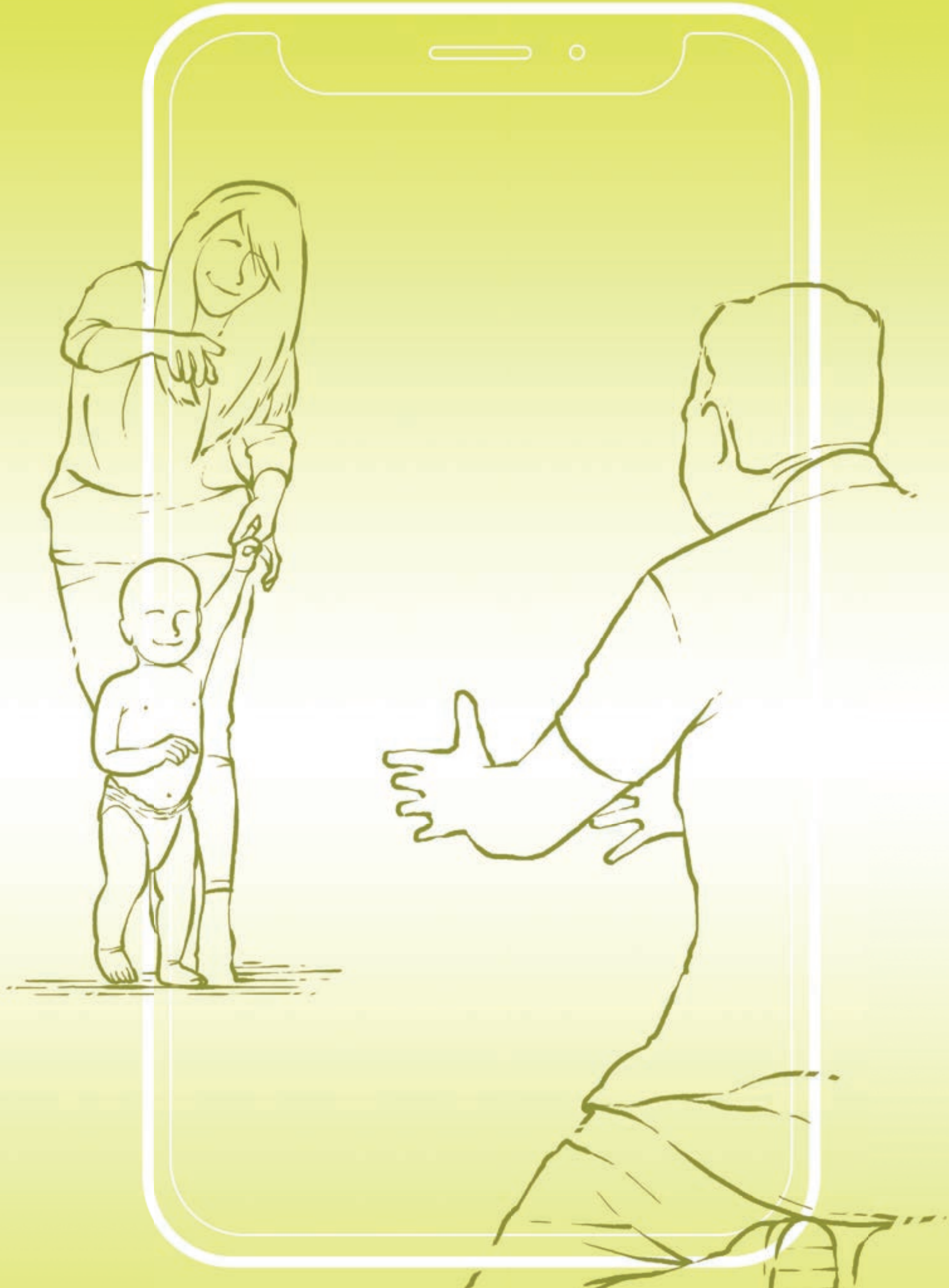
Following this, a quadratic model was built. This model includes, in addition to the intercept and linear slope previously described, also a quadratic slope, where the loadings quadratically increase. This model did converge, but model fit was insufficient ($\chi^2(12)$ = 22.68, *p* = .031, CFI = .97, TLI = .97, RMSEA = .09).

Next, a cubic model was built. This model included, in addition to the previously described intercept, linear and quadratic slope, also a cubic slope where the loadings increase cubically. This model failed to converge.

Finally, a sigmoidal model was built trying to model an S-shaped growth curve. To built this model we followed the syntax as suggested by Grimm and Ram (2009). This model also failed to converge. The repeated failure to converge was probably caused mainly by the sample size that is too small to fit such complex growth models. An additional issue is the smaller variance in the first and last measurement moments. However, models without these measurement points also did not converge, thus suggesting that the main problem is the sample size.

The only model that converged was the quadratic growth model. However, the model fit was still insufficient and a visual inspection of the data also suggests that quadratic growth does not do justice to the growth patterns seen in the data.

PART III



Chapter 5

Factors associated with gross motor development from birth to independent walking: A systematic review of longitudinal research

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ABSTRACT

Objective: To gain more insight into child and environmental factors that influence gross motor development (GMD) of healthy infants from birth until reaching the milestone of independent walking, based on longitudinal research.

Methods: A systematic search was conducted using Scopus, PsycINFO, MEDLINE and CINAHL to identify studies from inception to February 2020. Studies that investigated the association between child or environmental factors and infant GMD using longitudinal measurements of infant GMD were eligible. Two independent reviewers extracted key information and assessed risk of bias of the selected studies, using the Quality in Prognostic Studies tool (QUIPS). Strength of evidence (strong, moderate, limited, conflicting, no evidence) for the factors identified was described according to a previously established classification.

Results: In 36 studies, six child and 11 environmental factors were identified. Five studies were categorized as having low risk of bias. Strong evidence was found for the association between birthweight and GMD in healthy full-term and preterm infants. Moderate evidence was found for associations between gestational age and GMD, and sleeping position and GMD. There was conflicting evidence for associations between twinning and GMD, and breastfeeding and GMD. No evidence was found for an association between maternal postpartum depression and GMD. Evidence for the association of other factors with GMD was classified as 'limited' because each of these factors was examined in only one longitudinal study.

Conclusion: Infant GMD appears associated with two child factors (birthweight, gestational age), and one environmental factor (sleeping position). For the other factors identified in this review, insufficient evidence for an association with GMD was found. For those factors that were examined in only one longitudinal study, and are therefore classified as having limited evidence, more research would be needed to reach a conclusion.

INTRODUCTION

Infants show great variability in the attainment of the milestones of gross motor development. For example, independent walking is achieved between the ages of 8 and 17 months.[1] According to Dynamics Systems Theory, infant motor development emerges from the interaction between factors within the child and in the environment.[2] Therefore, many different factors are responsible for this variability in infant motor development.[3] Several studies have investigated the association between child factors and an infant's gross motor development (GMD). Some factors have been subjects of study in reviews including gestational age (GA) and birthweight (BW). In three reviews on these factors, strong evidence was found on lower outcomes on motor development in infants born very preterm or with a very low birthweight from birth till 16 years of age.[4–6] The review by Pin and colleagues,[7] about the factors sleeping position and the use of equipment, showed evidence for a transient delay in motor development of both term- and low risk preterm infants who were not exposed to prone position. The use of equipment does not seem to delay or speed up motor development in healthy term born infants. Reviews on other child and/or environmental factors are lacking. Furthermore, in the above-mentioned reviews, it was noted that many studies were of low methodological quality, and most included studies had a cross-sectional design. Because variability and time are key elements in GMD, studies with a repeated-measures design are preferred to those that evaluate the association of a factor cross-sectionally.[8] By examining the association between a factor and GMD over time using the same sample, findings based on sample differences are avoided. Hence, studies with longitudinal designs give a more reliable representation of factors associated with GMD than those with cross-sectional designs.[9]

A better understanding of factors associated with GMD of infants is an important basis for clinical reasoning and for designing new interventions for infants lagging in their GMD.[10] Given the small number of reviews on factors associated with GMD, their dates of publication, and the limited scope of factors included, it is important to provide an update. Besides, longitudinal studies relating to child factors and environmental factors associated with infant GMD have not yet been considered systematically. Therefore, the aim of the present review is to provide an overview of child and environmental factors associated with GMD of infants from birth to independent walking, based on longitudinal studies.

METHODS

Data sources and searches

A systematic search was conducted to identify studies that met the inclusion criteria. MEDLINE, CINAHL, PsycINFO and SCOPUS were searched from inception to February 2020. The search contained three main terms: 'motor development', 'infants' and 'cohort studies'. The search strategies, tailored to the different databases, are included in Appendix 5.1. When a systematic review was found, all included studies were screened for eligibility for this review.

Study selection

Only studies published in peer-reviewed journals in English, with full text available, were included. Two reviewers (IS, MB) selected the studies independently, first by title and abstract and then, if necessary, by reading the methods section of the study. If the reviewers could not reach consensus, a third independent reviewer (JN or MV) was consulted. All remaining studies were subsequently read in full text to determine eligibility according to inclusion and exclusion criteria.

For inclusion, a longitudinal design was required, meaning two or more repeated measurements of GMD. When the study outcome was the attainment of a motor milestone, only prospective parental reports were included. Participants had to be healthy preterm or full-term infants. Preterm infants with the following conditions were excluded: cystic periventricular leukomalacia; Grade III or IV hemorrhage according to Papile classification; post-hemorrhagic ventricular dilation; bronchopulmonary dysplasia (defined as oxygen supplementation > 36 weeks postmenstrual age). Studies on pathology or medical intervention were excluded. If no description of important characteristics such as gestational age, birth weight and the presence of pathology was available, the study was excluded. Only in birth cohort studies with samples that included > 1500 infants, a maximum of 5% percent of infants with health conditions that may affect motor development were accepted. At least one measurement of a child factor or an environmental factor, hypothesized to have an association with GMD, had to be reported. The following factors were excluded: prenatal factors (e.g., intra-uterine growth retardation) or specific maternal factors (e.g., drugs, intracytoplasmic injection) and interventions (e.g., zinc, baby massage).

Study quality/risk of bias

Critical appraisal of studies is essential to identify and assess biases that may have affected the study outcomes.[11] Therefore, two researchers (IS, MB) assessed all included studies (*n* = 36) independently with the Quality in Prognostic Studies tool (QUIPS). This tool is designed to assess the risk of bias (RoB) in studies with prognostic factors.[12] The QUIPS includes 31 questions on validity and bias in six areas: study participation, study attrition, prognostic factor measurement, outcome measurement, study confounding, and statistical analysis and reporting. The items are scored as "yes" (fulfilled), "partial" (partially fulfilled), "no" (not fulfilled), or "?" (unclear whether criterion is fulfilled). Subsequently, based on individual items' scores within each domain, all six domains were labelled "low", "moderate" or "high" RoB, according to the recommendations and prompts of Hayden et al.[12] Disagreement on individual scores was resolved by discussion and consensus. If necessary, a third reviewer (JN or MV) was consulted. Finally, a total RoB score was composed for each study as a basis for the best evidence synthesis. A study had to score a low RoB in all six domains for the overall RoB to be judged "low". If this requirement was not met, the study was rated as having a high overall RoB. This procedure was determined a priori by the reviewers and based on the procedure described by Hayden et al.[12] All information and discussion about RoB assessment is reported in Review Manager.[13] A summary statement of the study quality is displayed in the Results (Table 5.4).

Data extraction and data synthesis

The results were presented according to PRISMA guidelines.[14] Factors with statistical significance ($p < 0.05$) were reported for each study. Analyzing the data, it became evident that various types of analysis had been performed e.g., repeated-measures analysis, cross-sectional analysis, and analysis of the mean age of reaching milestones as outcome measure (motor milestone studies). Because these outcomes were so heterogeneous, a meta-analysis could not be conducted. Therefore, a qualitative synthesis was performed, and the strength of evidence assessed following the descriptions for prognostic studies according to Hayden et al. (2019), described in Table 5.1. Data extraction focused on population characteristics, ages and measurements for motor outcomes and factors. From the results, correlations, regression coefficients, odds ratios, and other outcomes were extracted (Supplementary Table S5.1).

Table 5.1 Strength of evidence (Hayden et al., 2019)

Strength of evidence	Description
Strong	Defined as greater than 75% of studies showing the same direction of effect in multiple low RoB studies
Moderate	Findings in multiple high RoB studies and/or 1 study with low RoB
Limited	1 study available
Conflicting	Inconsistent findings across studies
No evidence	No association between prognostic factor and outcome of interest

RESULTS

The search yielded 5594 potentially relevant studies. After removing duplicates, 3548 studies remained. These were screened independently by two reviewers on title and abstract and 3250 studies were excluded. Four studies were added from other sources. From the remaining 302 full text studies, 36 were eligible for this review. Reasons for exclusion are specified in the PRISMA flow chart[14] (Figure 5.1).

Study characteristics

Included studies had their origin in 13 countries. Of the 36 studies, 25 were conducted in North America and Europe, the others being mainly carried out in Asia (Taiwan and Japan) and South America (Brazil). In total, the studies represent 71546 infants with a median sample size of 261.5 [range 27–20,112]. In 22 of the included studies, only FT infants (GA ≥ 37 weeks) participated. Mixed populations (both full-term and preterm infants) were examined in 13 studies and one study included only preterm infants (GA < 34 weeks). Six child factors were examined in 16 studies and the association of 12 environmental factors was evaluated in 20 studies. The included studies table (Supplementary Table S5.1) provides information on the main population characteristics, study design, analyses performed, and outcomes. The studies were grouped by type of factor (child, environmental or multiple factors), see Table 5.3. Studies were described by the main factor, which was the main objective of the research question. Studies examining multiple factors were grouped. Confounders that were considered and were significant in the final model are summarized in the data extraction table (Supplementary Table S5.2). A summary of the significant associations of factors with GMD is displayed in Table 5.3.

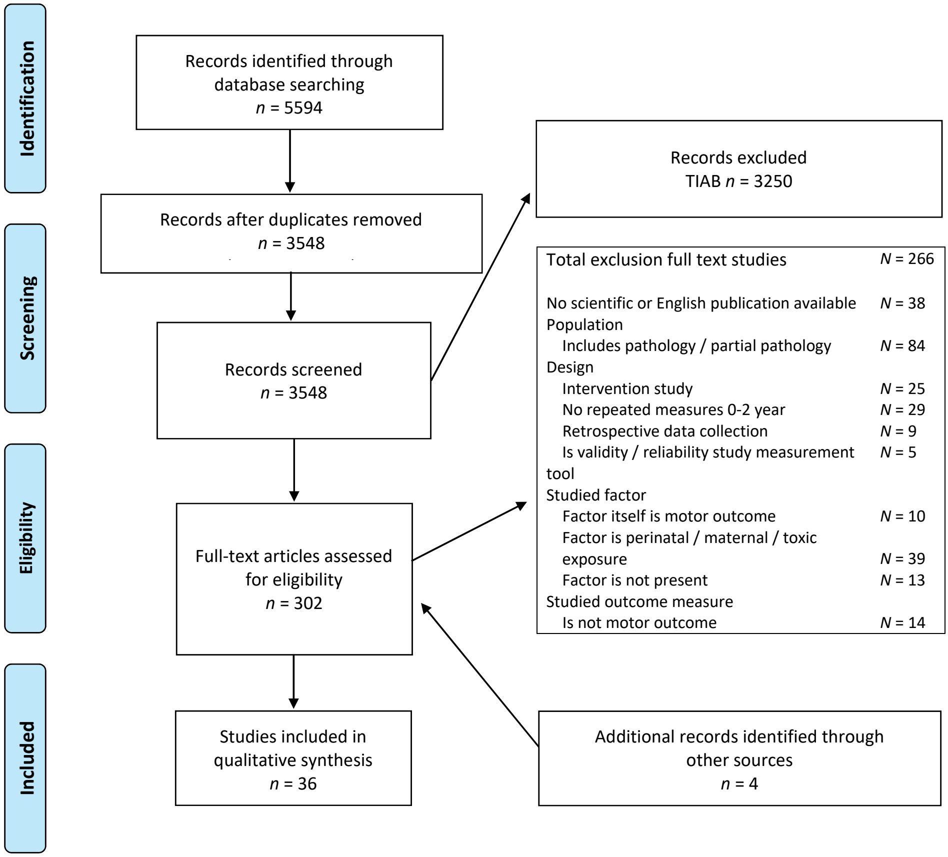

Figure 5.1 PRISMA flowchart.

Risk of bias assessment

Major issues with study quality were related to study attrition and study participation. High RoB on the domain 'statistical analysis and reporting' was mainly found in research carried out before the year 2000 (n = 4). Five studies scored an overall low RoB, comprising 14% of included studies.

Child factors

Gestational age

Four studies with high RoB examined the association of GA and GMD in various populations,[15–18] finding moderate evidence that a shorter GA for infants is negatively associated with GMD in the age range 0–18 months. The study by Yaari and colleagues[18] showed that moderately preterm (MPT) infants (GA 32–34 weeks) have persistently lower levels of GMD in the age range 1–18 months, compared to full-term infants. However, because GA and birthweight were highly correlated, it is not clear whether these differences are primarily due to GA or birthweight[17,18] concluded that most of the variance (14.5%) in the achievement of motor milestones by infants, both full-term and preterm, is explained by GA and birthweight. In a sample of full-term infants (37–41.6 weeks GA), longer pregnancy duration was also significantly associated with better motor scores at 3, 6 and 12 months, after adjusting for confounders.[15] There is no evidence for an association between GA in infants born post-term (> 42 weeks) and GMD from 4 to 12 months.[16]

Birthweight

Four studies examined the association between birthweight and infant GMD.[17,19–21] Two studies with a low RoB and one study with a high RoB examined infants with very low birthweight (VLBW) (< 1500 g) and found strong evidence that low birthweight (LBW) (< 2500 g) in both preterm and full-term infants is associated with a more delayed GMD in the age range 4–24 months.[19,20] There is limited evidence that infants with normal birthweight (> 2500 grams) have more advanced GMD than infants with LBW.[21] In a mixed population of infants (GA 27–46.5 weeks), Flensborg et al.[17] showed that birthweight in addition to GA explained most of the variance in motor milestone attainment. All studies that included preterm infants accounted their outcomes to GA.

Anthropometry

Three studies investigated the association of anthropometric measures with infant GMD. The study with the factor 'overweight'[22] had a low RoB; the other two had high RoBs.[23,24] Due to the heterogeneity of the populations and the difference in measures, the outcomes of these three studies could not be compared.

Regarding the factor 'overweight', there is moderately consistent evidence that overweight full-term infants, measured from birth to 18 months, are more prone to delayed GMD in the age range 3–18 months, compared with infants of normal weight.[22]

Limited evidence was found for the factors 'proportionately larger head', 'Body Mass Index (BMI)', and 'body length'. Infants with normal birthweight and a proportionately larger head showed lower motor scores at 6 weeks, but not at later ages.[23] For the factors 'body length' and 'BMI', no association was found with infant motor outcome between 6 weeks and 15 months.

For VLBW infants, there is limited evidence that BMI and length are associated with more delayed GMD at 9 and 24 months.[24]

Twin

Four studies with high RoB investigated the association between twinning and GMD, allowing for birthweight and GA.[25–28] Overall, the evidence was inconsistent: either significantly negative associations or no associations between GMD and twinning were reported. The study by Brouwer and colleagues[25] found no differences in the achievement of motor milestones between Dutch singletons and twins in the age range 0–24 months. Three other studies reported significant or non-significant associations at different ages. Nan et al.[27] reported that twins from 0 to 12 months scored lower on GMD, compared with singletons. These outcomes are broadly in line with the study by Goetghebuer et al.[26] After adjusting for the confounder birthweight, the age of milestone achievement was significantly greater for twins in only three out of eight milestones in the first year. Lastly, Wilson et al.[28] observed that twins had significantly lower motor scores compared to singletons at 6 and 18 months, but not at 3, 9 and 12 months.

Other child factors

For the child factors, 'Afro-American background' and 'motivation to move', significant associations with infant GMD were reported but, as each factor was examined by only one longitudinal study, each with high RoB, these findings were interpreted as providing limited evidence. Infants with an Afro-American background achieved most motor milestones at an earlier age compared to infants with other cultural backgrounds.[29] Infants that were perceived to a stronger motivation to move in the age range 7 to 12 months showed earlier achievement of five milestones.[30]

Environmental factors

Sleep position

In four studies, all high RoB, sleep position was examined in association with infant GMD. There is moderate evidence that prone sleeping is associated with a better GMD from 4 to 10 months.[31,32] No association was found from 11 to 17 months. In a study of Majnemer et al.,[32] prone-sleeping infants showed better GMD at 6 months, but not at 4 and 15 months. Davis et al.[31] showed an advantage for prone-sleeping infants in the attainment of several motor milestones in the range 4–10 months. There is no association between prone sleeping and the motor milestone 'walking alone'.

Table 5.2 Included studies and factors examined

Single factor studies																	
	Child factors						Environmental factors										
Author (year)	GA	BW	Twin	Anthro-pometry	Background	Motivation to move	Sleep position	Breast-feeding	Mat. depression	Mat. mental health	Adolescent mother	Par. mental health	Par.neo-natal perceptions	Baby walker	Day-care attendance	Season of birth	Cultural context
Yaari (2018)	X_c																
Espel (2014)	X_c																
Field (1978)	X																
Datar (2009)		X_c															
Grantham-McGregor (1998)		X_c															
Lung (2009)		X_c															
Nan (2013)			X_c														
Brouwer (2006)			X_c														
Goetghebuer (2003)			X_c														
Wilson (1975)			X_c														

	Child factors						Environmental factors										
Author (year)	GA	BW	Twin	Anthro-pometry	Background	Motivation to move	Sleep position	Breast-feeding	Mat. depression	Mat. mental health	Adolescent mother	Par. mental health	Par.neo-natal perceptions	Baby walker	Day-care attendance	Season of birth	Cultural context
Scharf (2016)				X_c													
Slining (2010)				X_c													
Bartlett (1998)				X													
Capute (1985)					X_c												
Atun-Einy (2013)						X											
Majnemer (2006)							X_c										
Davis 1998)							X_c										
Lung (2011)							X_c										
Ratliff (2001)							X_c										
Jardi (2017)								X_c									
Michels (2017)								X_c									

Table 5.2 continues on next page.

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Table 5.2 ***Continued***

Single factor studies																	
	Child factors						Environmental factors										
Author (year)	GA	BW	Twin	Anthro-pometry	Background	Motivation to move	Sleep position	Breast-feeding	Mat. depression	Mat. mental health	Adolescent mother	Par. mental health	Par.neo-natal perceptions	Baby walker	Day-care attendance	Season of birth	Cultural context
Morris (1999)								X_c									
Oddy (2011)								X_c									
Smith-Nielsen (2016)									X_c								
Sutter-Dallay (2011)									X_c								
Lung (2011)										X_c							
de Borba (2015)											X						
Lung (2009)												X_c					
Hernandez (2011)													X_c				
Siegel (1999)														X_c			
Souza (2010)															X		
Tsuchiya (2012)																X_c	
Vierhaus (2011)																	X

Multiple factor studies													
	Child factors						Environmental factors						
Author (year)	GA	BW	Sex	Weight increase	Larger head	Cognition	Days of mechanical ventilation	Breast-feeding	Higher birth order	Maternal age	Paternal age	Family income	More stimulating independent positions
Bjarnadottir (2019)	X		X					X		X			
Flensborg (2017)	X	X		X	X			X	X		X		
Pereira (2016)						X	X					X	X

5

Table 5.3 Associations between factors and infant GM

Child factors		Age of motor assessment outcome					Age of motor milestone outcome				
	RoB	0–3 m	4–6 m	7–12 m	13–24 m	25–60 m	0–3 m	4–6 m	7–12 m	13–24 m	25–60 m
GA											
Preterm											
Yaari, 2018	H			s**							
Term											
Espel, 2014	H	s*	s**	s**							
Postterm											
Field, 1978	H		ns	ns							
Birthweight											
Low BW											
Datar 2009	L			s***	s**						
Grantham Mc-Gregor, 1998	L		s***	s***							
Normal BW											
Lung, 2009	H		s***		s***						
Twin											
Nan, 2013	H	s***	s***	s***	ns						
De Brouwer, 2006	H								ns	ns	
Goetgebuer, 2003	H						s**	s*/ns§	s*/ns§		
Wilson 2006	H	ns	s***	ns	s***						

		Age of motor assessment outcome					Age of motor milestone outcome				
	RoB	0–3 m	4–6 m	7–12 m	13–24 m	25–60 m	0–3 m	4–6 m	7–12 m	13–24 m	25–60 m
Anthropometry											
Weight, length, head circumference											
Scharf, 2018	H		s***		s***						
Overweight											
Slining, 2010	L	s*									
Proportionately larger head											
Bartlett, 1998	H	ns/s***	ns	ns	ns						
Afro-American background											
Capute, 1985	H						s**	s**	s**	s**	
Motivation to move											
Atun-Einy, 2013	H			s*					s*/**/**		

Table 5.3 continues on next page.

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Table 5.3 ***Continued***

Environmental factors		Age of motor assessment outcome					Age of motor milestone outcome				
	RoB	0–3 m	4–6 m	7–12 m	13–24 m	25–60 m	0–3 m	4–6 m	7–12 m	13–24 m	25–60 m
Sleep position											
Prone sleeping											
Majnemer, 2006	H		ns∞/s∞		ns						
Davis, 1998	H							$s^{**}/^{**}/^{***}$/ns	$s^{**}/^{**}$/ns		
Supine sleeping											
Lung, 2011	H		s^{***}		ns	ns					
Ratliff, 2001	H		ns		ns						
Breastfeeding											
Jardi, 2017	L		s^{*}	s^{*}							
Michels, 2017	H		ns	ns	ns						
Morris, 1999	L		ns/$s_{LBW/HBW*}$	ns							
Oddy, 2011	H			ns	ns	ns					
Maternal depression											
Smith-Nielsen, 2019	H		ns		ns						
Sutter-Dallay, 2011	H	ns	ns	ns	ns						
Maternal mental health											
Lung, 2010	H		ns		ns	ns					
Adolescent mother											
de Borba, 2015	H		ns								
Parental mental health											
Lung, 2009	H		ns		$s_{m}*$						

		Age of motor assessment outcome					Age of motor milestone outcome				
	RoB	0–3 m	4–6 m	7–12 m	13–24 m	25–60 m	0–3 m	4–6 m	7–12 m	13–24 m	25–60 m
Parental neonatal perception											
Hernandez, 2011	H		s_m*	s_p**							
Baby walker											
Siegel, 1999	H			s ***				s***/**	s**		
Daycare attendance											
Souza, 2010	H			s	s						
Season of birth winter											
Tsuchiya, 2012	H		s***	s***	ns						
Cameroonian vs German culture											
Vierhaus, 2011	H		s ns								

Multiple factors

		Age of motor assessment outcome					Age of motor milestone outcome				
	RoB	0–3 m	4–6 m	7–12 m	13–24 m	25–60 m	0–3 m	4–6 m	7–12 m	13–24 m	25–60 m
Bjarnadottir, 2019	H						ns	ns	ns	ns	
Pereira, 2016	H		s								
Flensborg, 2017	H						s***	s***	s***		

Note: L = low RoB (Risk of Bias), H = high RoB; LBW = low birthweight, HBW = high birthweight; ns = no significant association, s = significant association; * p < .05, ** p < .01, *** p < .001; s/ns = (no) associations longitudinally analyzed; m = maternal, p = paternal; § = multiple motor milestones measured in same age-range; ∞ = multiple motor outcomes measured in same age-range.

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Table 5.4 Risk of bias assessment (QUIPS)

	Study participation	Study attrition	Prognostic factor measurement	Outcome measurement	Study confounding	Statistical analysis and reporting	Overall RoB
			Child factors				
GA							
Preterm							
Yaari, 2018	?	+	+	?	+	+	H
Term							
Espel, 2014	+	?	?	+	+	+	H
Postterm							
Field, 1978	?	N/A	-	+	-	-	H
Birthweight							
Low birthweight							
Datar, 2009	+	+	+	+	+	+	L
Grantham Mc-Gregor, 1998	+	+	+	+	+	+	L
Normal birthweight							
Lung, 2009	+	+	+	?	+	+	H
Twin							
Nan, 2013	+	-	+	?	+	+	H
Brouwer, 2006	?	N/A	+	?	?	-	H
Goetgebuer, 2003	?	-	+	?	+	+	H
Wilson 2006	-	-	-	+	-	-	H
Weight, length, head circumference							
Scharf, 2018	?	?	+	+	+	?	H
Overweight							
Slining, 2010	+	+	+	+	+	+	L

	Study participation	Study attrition	Prognostic factor measurement	Outcome measurement	Study confounding	Statistical analysis and reporting	Overall RoB
Proportionately larger head							
Bartlett, 1998	+	-	+	+	-	-	H
Afro-American background							
Capute, 1985	?	-	?	-	-	-	H
Motivation to move							
Atun-Einy, 2013	-	N/A	-	+	-	?	H
Environmental factors							
Sleep position							
Prone sleeping							
Majnemer, 2006	-	?	?	+	+	+	H
Davis, 1998	+	?	+	?	?	+	H
Supine sleeping							
Lung, 2011	?	?	+	?	+	+	H
Ratliff, 2001	?	-	?	+	+	-	H
Breastfeeding							
Jardi, 2017	+	N/A	+	+	+	+	L
Michels, 2017	+	N/A	+	?	+	+	H
Morris, 1999	+	+	+	+	+	+	L
Oddy, 2011	?	?	?	?	+	+	H
Maternal depression							
Smith-Nielsen, 2019	?	+	+	+	?	+	H
Sutter-Dallay, 2011	+	?	?	+	+	+	H

Table 5.4 continues on next page.

Table 5.4 ***Continued***

	Study participation	Study attrition	Prognostic factor measurement	Outcome measurement	Study confounding	Statistical analysis and reporting	Overall RoB
Maternal mental health							
Lung, 2011	+	?	+	?	+	+	H
Adolescent mother							
de Borba, 2015	-	N/A	+	-	?	-	H
Parental mental health							
Lung, 2009	+	+	+	?	+	+	H
Parental neonatal perception							
Hernandez, 2011	+	N/A	+	+	+	?	H
Baby walker							
Siegel, 1999	?	+	?	+	?	?	H
Daycare attendance							
Souza, 2010	-	N/A	?	+	-	-	H
Season of birth winter							
Tsuchiya, 2012	?	+	+	?	+	+	H
Cameroonian vs German culture							
Vierhaus, 2011	?	?	+	?	+	+	H
Multiple factors							
Bjarnadottir, 2019	+	+	+	?	+	+	H
Flensborg, 2017	+	-	+	?	+	?	H
Pereira, 2016	-	-	?	+	+	+	H

Note: L = low RoB (risk of bias), H = high RoB; - = not reported, + = reported, ? = not sure; N/A = not applicable.

Conflicting evidence is found for the association between supine sleeping and a lower score on GMD at 4 and 6 months.[33,34] No evidence was found associating supine sleeping with GMD in the age ranges 0–3 and 12–36 months. In a cohort study of Lung et al.,[33] supine-sleeping infants showed a delay in GMD at 6 months: at 18 and 36 months, the association was no longer present. Ratliff et al.[34] studied a population of very preterm (VPT) infants. GMD at the corrected ages of 4 and 13 months was not associated with sleeping supine.

Breastfeeding

Five studies, two with low RoB[35,36] and three with high,[37–39] investigated the association between breastfeeding and infant GMD. Two studies had mixed populations (preterm/full-term infants and LBW/HBW full-term infants), one was a cohort study, and two studies examined full-term infants. Breastfeeding duration as a factor was defined differently in all studies and, overall, conflicting evidence was found regarding the role of breastfeeding. Jardi et al.[35] reported, in a low RoB study, a significant association of exclusive breastfeeding and mixed feeding till 4 months with advanced GMD at 6 months in full-term infants as compared to infants who received only formula feeding. These associations were only significant in the adjusted model when the factors BMI at 6 months and GA were added. At 12 months, a significant association of exclusive breastfeeding with advanced GMD was present when the factor iron status was added to the model.

In four studies, no evidence was found of an association between breastfeeding and GMD in the first three years of life in diverse populations.[36,38,39] Morris et al.,[38] a low RoB study, compared groups of full-term infants with HBW and LBW and evaluated the frequency of breastfeeding in the first 4 weeks and between 5 and 26 weeks. They found that breastfeeding intensity did not correlate with motor outcome at 6 and 12 months for both groups separately. Linear regression showed that in both LBW and HBW infants, breastfeeding intensity in the first 4 weeks of life was significantly associated with motor scores at 6 months but this was no longer apparent at 12 months.[38] Michels et al.[36] did not find an association of exclusive breastfeeding and infant GMD, nor for preterm infants. The study by Oddy et al.[39] revealed that GMD scores in infants with breastfeeding < 4 months did not differ from those in infants with breastfeeding> 4 months. Only boys who were breastfed for less than 4 months had an increased risk of one atypical score on the Ages and Stages Questionnaire (ASQ) at any time-point. In the group of full-term infants with normal birthweight, Bjarnadóttir et al.[37] found no association between duration of breastfeeding (exclusive or total duration) and motor milestone achievement.

Maternal depression

In two studies, both with high RoB, maternal depression was examined in association with infant GMD. Overall, there is no evidence that postpartum depression is associated with GMD between the ages of 3 and 24 months.[40,41] In the study of Smith-Nielsen,[40] 28 full-term infants of mothers with a diagnosis of maternal depression were compared to a control group (*n* = 53). This revealed no association with motor scores at the ages of 4 and 13 months. Sutter-Dallay et al.[41] found no association between the depression score of the mother (at six weeks after giving birth and at follow-up) and GMD from 3 to 24 months.[41]

Other environmental factors

The following environmental factors were examined by only one longitudinal study each and findings are therefore categorized as high RoB, interpreted as limited evidence.

For the environmental factors 'use of an occluding baby walker', 'home environment' and 'daycare attendance', significant associations with infant GMD were reported. The use of an occluding baby walker, a walker in which the infant is not able to see its own feet, is significantly associated with delayed GMD between 6 and 15 months, in comparison to a see-feet baby walker and no baby walker use.[42] Home environment, including higher family income, more stimulation and putting the infant in independent positions, is significantly associated with higher motor performance in infants between 2 and 12 months.[43] For daycare attendance, it was found that, of infants attending full-time, 13% (*n* = 4) had suspected motor delays at 12 and 17 months.[44]

For the factors each examined by one high RoB study, season of birth,[45] parental mental health,[46] parental neonatal perceptions,[47] and cultural context,[48] the association with GMD changed over time. Infants born in spring have higher motor scores at 6 and 10 months than infants born in winter; at 14 months, no association with GMD is found.[45] Better parental mental health is associated with better GMD at 18 months.[46] Concerning the factor 'parental neonatal perceptions', more negative maternal perceptions have a negative association with infant GMD at 4 months. At 12 months, positive paternal perceptions were associated with an advanced GMD.[47] Cameroonian infants have significantly higher motor scores than German infants at 3 and 6 months, implying an association between cultural context and GMD. At 9 months, this association was no longer present.[48]

No evidence was found for the factor 'adolescent mother'. Motor scores of infants aged 0 to 18 months did not differ significantly whether they had adolescent or adult mothers.[49]

DISCUSSION

This review aimed to provide an overview of factors associated with GMD of healthy full-term and preterm infants as examined in longitudinal studies. In total, 36 studies were identified of which 15 examined a child factor, 17 examined an environmental factor and 4 investigated multiple factors. Six child factors and 11 environmental factors were examined in the selected studies. Strong evidence was found for the association of the child factor 'LBW' with infant GMD. Moderate evidence was found for the child factors 'overweight' and 'shorter GA', and for the environmental factor 'prone sleeping'. There was conflicting evidence for the factors 'twinning', 'supine sleeping' and 'breastfeeding'. Regarding the other factors identified in this review, insufficient evidence for an association with GMD was found and they were classified as having no or limited evidence. Only the factors which are examined in multiple studies and therefore enabling a qualitative synthesis will be discussed in more depth.

Child factors

This review included four longitudinal studies,[15–17,24] all showing moderate evidence that a shorter GA is associated with a delay in GMD. The samples that were studied ranged from 26 to 42 weeks GA. This association is in line with the results from the meta-analysis in the review by de Kieviet et al.[4] who reported a significant negative association between the GA of VPT children and GMD. The study of Espel et al.[15] indicated that the duration of gestation is not only associated with GMD in preterm infants but also, maybe less pronounced, in early full-term, full-term and late full-term infants. Fundamentals about the association of gestational age with GMD presented in most of the included studies[15,18] are that growth of the brain and neurological maturation of the brain during the prenatal period are linked to neurodevelopmental outcome.

This review provides strong evidence that both VLBW (< 1500 g) and LBW (1500–2500 g) are significantly associated with lower motor outcomes of preterm and full-term infants from 0 to 24 months. These findings concur with those of a systematic review on motor outcomes in VLBW and VPT children,[4] including a meta-analysis on 9653 VLBW children from 0 to 16 years. De Kievit et al.[4] concluded that an increase in birthweight related to better GMD. The negative association of LBW and GMD was also reported in a cross-sectional study of Hediger,[50] who found delays in GMD in both full-term and preterm infants with LBW. These outcomes show that the impact of birthweight on GMD transcends that of premature birth. Golding et al.[6] concluded that LBW is a marker of intra-uterine growth retardation rather than of preterm delivery and therefore has a direct and strong impact on GMD. From the included studies, only the study of Datar and Jacknowitz[19] provides an explanation of the relation between birthweight and GMD. Not only intrauterine malnutrition but also genetic and/or environmental effects may cause low birthweight and therefore a lower GMD outcome in the first years of life.[19]

Regarding the factor 'twinning', it is known that twins are more prone to developmental delay from prematurity and LBW. The question arises of whether twinning is an independent risk factor. In this review, conflicting evidence was found in four studies.[25–28] Differences in the sample and in the method of measuring GMD might play a role in this. Goetghebuer et al.[26] found that Gambian twins were significantly delayed in reaching three of the eight milestones studied, after adjustment for the confounders birthweight and GA. However, the authors suggest that cultural factors may explain the observed delays in the twins' GMD. In the Dutch sample of Brouwer et al.,[25] no significant differences were observed in GMD between twins and singletons with normal birthweight and GA. Unlike the study of Goetghebuer et al.,[26] who used the mean age of reaching a milestone, Brouwer et al.[25] used the percentage of twins who achieved a milestone at a fixed age, which is less accurate and might explain differences in outcomes. A study performed in the United Kingdom (UK) measured GMD of infants (GA 26–39 weeks), using the ASQ, and based the outcomes on the American norm scores of healthy full-term singletons.[27] This study found that UK twins scored below the normal range on GMD until 9 months of age. However, a singleton control group was not used. Recent research on the cross-cultural validity of norm values of motor measurements shows that North American infants are ahead of European infants.[51–53] In this light, it might be debated whether the described results are indicators of delayed GMD in twins or merely a reflection of normal GMD in UK preterm and full-term infants. Overall, the evidence from these longitudinal studies does not show that twinning is an independent risk factor for GMD of infants.

Environmental factors

The included studies on the factor breastfeeding, all provide equal hypotheses about why GMD may be positively affected by breastfeeding, namely 1) breastfeeding is a critical source of energy enabling motor development and, 2) breastfeeding protects infants against gastrointestinal infections which optimizes health and therefore (motor) development. In this review, no evidence of an association between breastfeeding and GMD was found in four studies.[36–39] This is in line with recent cross-sectional studies,[54,55] and a review by Golding et al.[6] which included six cross-sectional studies and also found no clear evidence for any association of breastfeeding with GMD. Despite these unequivocal findings, Jardí et al.[35] found a positive association between breastfeeding and GMD in a group of term born infants with a normal birthweight that were exclusively breastfed at the age between 6 and 12 months and received mixed feeding at the age of 4 months. The outcomes were only significant in the adjusted model including GA and BMI at 6 months and iron status at 12 months. This might indicate that any existing relationship between breastfeeding and GMD is mainly indirect and based on infant anthropometry and important nutrients like iron. Considering the limitations that are mentioned in the included studies, it becomes evident that rigorous research in this field is a challenge. One reason for this is the many confounding factors, such as maternal cognition and socio-economic effects. Besides, the effects of breastfeeding appear to be different in developing and developed countries and in term born and preterm born infants with a low birthweight. Finally, several studies report that the lack of an association between GMD and breastfeeding might also be due to the formula feeding that improved so much over the last decades that it levels the quality of breastmilk[36,37,39] concludes that the positive effects of breastfeeding go beyond motor development.

The moderate evidence found in this review for a positive association of prone sleeping and GMD from 4 to 10 months for both full-term and preterm infants was already signaled in the review of Pin et al. which included nine studies on the effects of sleeping position on GMD.[7] Three of these studies were longitudinal and are included in this review.[31,32,34] The study of Lung et al.[33] concluded that supine sleepers only showed a delayed GMD at 6 months, not at 18 and 36 months. It seems logical that the association between sleeping position and GMD is most present before 6 months when infants are dependent on their caregivers to change positions. There are also indications that more than 20 years after the ‘Back to sleep’ campaign was set up, the adverse effects on GMD of supine sleeping might have diminished due to more adequate education about ‘tummy time’.[56,57]

There was no evidence found in the two included studies for an association between postpartum maternal depression (PPMD) and GMD in infants.[40,41] A systematic review of nine studies by Aoyagi et al.,[58] including the study of Smith-Nielsen, also found no association between GMD and PPMD. The studies of Smith-Nielsen et al.[40] and Sutter-Dallay et al.[41] do both not explain the mechanism that links PPMD to delayed motor development. Regarding the other environmental factors which were examined in single studies with a high RoB, only the effect of baby walker use on GMD has been previously reviewed.[7,59] The cohort study of Siegel & Burton,[42] included in this review, was included in both reviews. Pin et al.[7] reported conflicting evidence; Burrows and Griffiths[59] conducted a pooled analysis of four studies and found a delay of 11 to 26 days in the onset of walking for infants using an occluded baby walker, which is in line with the outcome of the study of Siegel & Burton.[42] Both reviews evaluated overall study quality as poor.

Strengths and limitations

In 18 of 36 studies, mean birthweight and mean GA were not reported. The absence of these major characteristics made comparisons difficult. Furthermore, the characteristics of the samples varied between studies examining the same factor. This heterogeneity in population characteristics improves the generalizability of the outcomes found in this review. In addition, the QUIPS has proved to be a useful tool to assess the quality of observational studies. This approach is supported by Huguet et al.[11] who, in addition advocate the use of modified GRADE standards to judge the quality of prognosis studies.

Future directions

In this review, inadequate study participation, high attrition and the lack of some robust measures for environmental factors seem to be the main causes of low study quality. Therefore, more high-quality studies need to be performed and replicated in the field to increase the levels of evidence.

In future research, using clearly described population groups, a fixed set of confounders and measures regarding infant GMD would enable researchers to draw more firm conclusions. Results from this review suggest that birthweight and GA should be considered as confounders for their profound impact on GMD.

To increase the number of longitudinal studies including large cohorts of infants, feasibility should be improved by lowering the burden for both infants and parents in time and costs. Innovative and digital aids, like smartphone apps and activity trackers, are possible means for gathering large amounts of data to provide insight into the complex pathways of infant development.[60–62] Also, more robust measures for environmental factors, like the home situation, caregiving practices and parent-infant interaction, are needed. Outcomes of these ‘modifiable factors’ can be the building blocks in developing new effective interventions to improve infant GMD.[10]

To date, evidence reveals that lower birthweight and shorter GA have a persisting negative association with GMD of infants over time. For many other factors, the association with GMD remains unclear. Overall, it can be concluded that our knowledge on what drives motor development in infants is still limited. To disentangle the complex interplay of genetic and environmental factors and their association with GMD, more research is needed.

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Key messages

- Low birthweight and short gestational age have a persisting negative association with infant gross motor development from birth to independent walking.
- There is inconsistent evidence for an association of breastfeeding, supine sleeping and (occluded) baby-walker use with infant gross motor development.
- More robust measures for environmental factors are needed.

Author contributions

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Supplementary Table S5.1 Characteristics and results of the included studies

Author (year, country)	Number of participants and participant characteristics	Motor measures and ages at measurement	Factor measures and ages at measurement	Confounders (*in the final model)	Statistical analysis and results
Yaari (2018, Israel)	*n* = 149 Groups: FT *n* = 39 *M* GA = 39.8 weeks (*SD* = 1.0) (range = 37.7–41.2) *M* BW = 3373 g (*SD* = 346) MPT *n* = 57 *M* GA = 33.2 weeks (*SD* = 0.6) (range = 32.1–34) *M* BW = 1865 g (*SD* = 320) VPT *n* = 34 (NI) EPT *n* = 19 (NI)	**Motor:** MSEL **Age:** 1, 4, 8, 12 and 18 months	**Factor:** GA **Measurement:** medical status FT (GA 37–41 weeks, BW > 2500 g) MPT (GA 33–34 weeks, BW < 2500 g) **Age:** at birth	Sex*	Regression with pairwise comparisons show that average level of gross motor outcome across time (from 1 to 18 months) is lower for MPT than for FT infants (b^* = -2.19, *SD* = 1.09, p = 0.045). Pairwise comparisons between MPT and FT on differences between gross motor outcomes between 18 months and one month, shows that MPT are more delayed in GMD than FT infants (b^* = -6.60, *SD* = 2.23, p = 0.0036).
Espel (2014, USA)	*n* = 232 Groups: Early term: 27% Full term: 56% Late term: 6.6% *M* GA = 39.46 weeks (*SD* = 1.06) (range = 37 0/7 – 41 6/7) *M* BW = 3418 g (*SD* = 420)	**Motor:** BSID- II **Age:** 3, 6 12 months	**Factor:** GA **Measurement:** ultrasound < 20 weeks of gestation Early FT (37–38 weeks) FT (39–40 weeks) Late FT (41–42 weeks) **Age:** at birth	BW GA Sex Birth order Ethnicity*	ANCOVA reveals group differences in psychomotor development at each assessment age. At three months, PDI is lower for early FT infants than for late FT ($F(2, 179)$ 54.01, p < 0.05). Early FT infants exhibit lower psychomotor development scores than FT and late FT infants at 6 months ($F(2, 168)$ 56.69, p < 0.01) and 12 months ($F(2,155)$ 55.32, p < 0.01). FT infants had lower psychomotor development scores than late FT infants at 12 months.

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Supplementary Table S5.1 ***Continued***

Author (year, country)	Number of participants and participant characteristics	Motor measures and ages at measurement	Factor measures and ages at measurement	Confounders (*in the final model)	Statistical analysis and results
Field (1978, USA)	*n* = 151 Groups: Post Term *n* = 46 *M* GA = 42 weeks *M* BW = 3600 g FT *n* = 59 *M* GA = 40 weeks *M* BW: 3300 g Post Term RDS *n* = 46 (NI)	**Motor:** DDST, BSID **Age:** 4, 8, 12 months	**Factor:** GA Measurement: N/A **Age:** at birth	No confounders considered	MANOVA showed that, at 4 months, post FT infants had inferior ratings on the DDST in comparison to the normal FT infants ($p < 0.001$). At 8 and 12 months, there were no significant differences between post FT and FT infants on the PDI scores.
Datar (2009, USA)	*n* = 7425 Groups: Singletons *n* = 6750 Twin pairs *n* = 625 Twins and other higher births whose siblings not included *n* = 50 *M* GA = 38.3 weeks *M* BW not reported	**Motor:** BSID-II SF **Age:** 9, 24 months	**Factor:** BW VLBW < 1500 g MLBW 1500–2499 g NBW ≥ 2500 g **Measurement:** weight **Age:** at birth	BW GA* Sex Birth order* Height* Ethnicity* Education* Income Marital status* Pregnancy/delivery risk factors*	At 9 months, multiple linear regression revealed large and significant effects of VLBW ($b^* = -8{,}764$; $p < 0.001$) and MLBW ($b^* = -2{,}901$; $p < 0.001$) on GMD. At 2 years, the cross-sectional estimates of VLBW ($b^* = -4.123$; $p < 0.001$) and MLBW ($b^* = -1.383$; $p < 0.001$) were considerably smaller, these changes being statistically significant at $\alpha = 0.01$. This suggests some catch-up is taking place between LBW and NBW children by age of two years.
Grantham-McGregor (1998, Brazil)	*n* = 262 Groups: ABW *n* = 131 LBW *n* = 131 GA > 37 weeks *M* GA and *M* BW not reported	**Motor:** BSID **Age:** 6, 12 months	**Factor:** BW ABW 3000–3499 g LBW 1500–2499 g **Measurement:** weight **Age:** at birth	SES	At 6 months, multiple linear regression showed that LBW-FT infants have significantly lower scores than ABW infants on the PDI (-7.3 points, $p < 0.001$). This difference increased by 12 months of age (PDI -9.9 points, $p < 0.001$).

Lung (2009, Taiwan)	*n* = 20112 Groups: FT *n* = GA ≥ 37 weeks BW ≥ 2500 g PT *n* = GA < 37 weeks BW < 2500 g *M* GA and *M* BW not reported	**Motor:** TBCS **Age:** 6, 18 months	**Factor:** twin, BW **Measurement:** N/A **Age:** at birth	BW* GA* Sex* Twin* Maternal education* Parental income*	Using structural equation modelling at 6 months, infants of parents with a higher income and infants born FT or with normal BW showed advanced GMD (b^* = 0.03, p < 0.001; b^* = - 0.11, p < 0.001; b^* = -0.10, p < 0.001). At 18 months, infants of mothers with a higher education, and of parents with higher income, who were male, twin, born FT of normal BW, had better GMD (b^* = 0.03, p < 0.001; b^* = 0.06, p < 0.001; b^* = 0.02, p = 0.019; b^* = -0.02, p = 0.026; b^* = -0.02, p = 0.036; b^* = -0.05, p < 0.001). (Model with p = 0.227 and AGFI = 0.999).
Nan (2013, UK)	*n* = 152 Twins *M* GA = 37 weeks (range = 26–39) *M* BW = 2300 g (range = 940–3500)	**Motor:** ASQ **Age:** 3, 6, 9, 12, 18, 24 months	**Factor:** twin, BW **Measurement:** birth chart **Age:** at birth	GA* BW Sex*	Cross-sectional multilevel linear regression analysis adjusted for sex and GA showed that twins scored lower on GMD than singletons (p < 0.001) during the first year of life. After the age of 12 months, twins catch up on GMD. BW was not a significant predictor of GMD at any age of measurement.
Brouwer (2006, The Netherlands)	*n* = 3490 Groups: Monozygotic twins *n* = 786 Dizygotic twins *n* = 1645 Singletons *n* = 1059 GA > 36.5 weeks BW > 2500 g *M* GA and *M* BW not reported	**Motor:** MM 4 milestones: turn, sit, crawl and walk **Age:** 0–24 months	**Factor:** twin **Measurement:** questionnaires, blood typing **Age:** ≥ 3 years	GA* BW (highly correlated with GA)	ANOVA shows that no remarkable differences are seen between healthy singletons and healthy twins in the achievement of gross motor milestones within the normal range. Dizygotic twins were faster than monozygotic twins in reaching the moment for sit (p < 0.001), crawl (p = 0.013), stand (p < 0.001) and walk (p < 0.001).

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Supplementary Table S5.1 ***Continued***

Author (year, country)	Number of participants and participant characteristics	Motor measures and ages at measurement	Factor measures and ages at measurement	Confounders (*in the final model)	Statistical analysis and results
Goetghebuer (2003, UK)	n = 408 Groups: Twin pairs n = 168 *M* GA twins = 38.9 weeks (range = 38.7–39.2) *M* BW twins = 2790 g (range = 2700–2800) Singletons n = 72 BW > 2500 g *M* GA singletons = 38.3 weeks (range = 38.1–38.6) *M* BW = 3240 g (range = 3100–3300)	**Motor:** MM 8 milestones adapted from DDST **Age:** 1, 2, 3, 4, 5, 9, 12, 18 months	**Factor:** twin **Measurement:** twin delivery **Age:** at birth	BW* Number of siblings* Non-independence within twin pairs* Length*	Age of milestone achievement was higher in twins for each milestone and significant for: Maintaining head (p = 0.003), Sitting without support (p = 0.03), Walking (holding on) (p = 0.03). Age of milestone achievement was highly concordant within twins. The concordance was significantly higher (p < 0.05) in monozygotic than in dizygotic twins for crawling, sitting, standing holding on, and taking 2 steps. At 12 months, after adjustment for BW, length and sex, twin status and number of siblings were significantly associated with 'parental report infant shows slower development than siblings' (p = 0.05) and 'maintaining head' (p = 0.05).
Wilson (1975, USA)	n = 261 *M* GA and *M* BW not reported	**Motor:** BSID **Age:** 3, 6, 9, 12, 18 months	**Factor:** twin **Measurement:** blood typing **Age:** at birth	BW*	Correlations show that twins have significantly lower scores on the motor scale at 6 and 18 months. Low GA in twins has a major effect on developmental status in the first half year of life (correlations at 3, 6, 9, and 12 months r = 0.30, r = 0.40, r = 0.20, r = 0.20, by 18- and 24-months p < .001).
Scharf (2016, USA)	n = 950 GA: ≥ 37 weeks = 3% 32–37 weeks = 18% 28–<32 weeks = 46% 22–<28 weeks = 34% VLBW: < 1500 g Groups: Anthropometric scores < -2 *SD* Anthropometric scores > 2 *SD*	**Motor:** BSID SF **Age:** 9, 24 months	**Factor:** weight, length, head circumference **Measurement:** weight **Age:** at birth, 9, 24 months	BW* Sex GA	Linear regression analysis adjusted for BW, sex and SES show that length and weight z-scores at 9 months were correlated with 1) children's Bayley motor scores at 2 years, and 2) the change in Bayley motor scores from 9 to 24 months. Children who scored more than 2 *SD*s below the mean in weight at 9 months showed a significant odds ratio (OR 2.64, p < .01) for Bayley motor scores of 2 SDs below the mean at 2 years.

Slining (2010, USA)	*n* = 217 GA > 35 weeks *M* GA = 39.48 weeks (*SD* = 1.47) *M* BW = 3.23 g (*SD* = 0.48)	**Motor:** BSID-II **Age:** 3, 6, 9, 12, 18 months	**Factor: weight** **Measurement:** weight and subcutaneous fat **Age:** at birth, 3, 6, 9, 12, 18 months	Age* Age squared* Sex* Weight status	Multivariate models showed that motor delay is 1.80 times more likely in overweight infants compared with non-overweight infants (i.e., weight-for-length *z*-score > 90th percentile) (95% CI [1.09, 2.97]) and 2.32 times as likely in infants with high subcutaneous fat compared with infants with lower subcutaneous fat (95% CI [1.26, 4.29]). High subcutaneous fat was also associated with delay in motor development (OR 2.27, 95% CI [1.08, 4.76]).
Bartlett (1998, Canada)	*n* = 132 BW > 2500 g *M* GA and *M* BW not reported	**Motor:** AIMS, PDMS **Age:** 6 weeks, 3, 5, 7, 10, 15 months	**Factor:** head proportion, BMI and body length **Measurement:** standard anthropometric measurements **Age:** 6 weeks, 3, 5, 7, 10 and 15 months	No confounders considered	Pearson correlations between head proportion and AIMS total, and subscale scores, revealed that infants with proportionately larger heads had significant lower scores on the AIMS total ($r = -0.38$, $p = 0.001$), this outcome being fully explained by the prone motor scores at 6 weeks of age. There was no correlation between BMI and body length and motor outcome scores.
Capute (1985, USA)	*n* = 381 *M* GA and *M* BW not reported	**Motor:** MM 12 motor milestones **Age:** N/A	**Factor:** ethnicity **Measurement:** N/A **Age:** time of recruitment	Sex SES	Analysis of variances show that infants with an Afro-American background achieve motor milestones, on average, at an earlier age, except 'Roll prone to supine'. Between 4 and 5 months of age, the milestones 'Roll supine to prone' was reached 0.5 month earlier by infants with Afro-American background. This advantage increases to 1.1 months for the milestone 'walk' (10.9 months vs 12 months). Association of ethnicity with motor gradient without adjustment is F(16.88 $p < 0.01$)). After adjusting for SES and sex, the association of ethnicity still exceeds $p < 0.01$-level.

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Supplementary Table S5.1 ***Continued***

Author (year, country)	Number of participants and participant characteristics	Motor measures and ages at measurement	Factor measures and ages at measurement	Confounders (*in the final model)	Statistical analysis and results
Atun-Einy (2013, Israel)	*n* = 27 *M* GA and *M* BW not reported	**Motor:** AIMS (video) **Age:** 7–12 months / every 3 weeks	**Factor:** MTM **Measurement:** MTM scale **Age:** 7–12 months / every 3 weeks. 7 measurements	No confounders considered	A repeated-measures ANOVA on the MTM score over the course of the 7 observations reveals a main effect for the group: $F(1, 18) = 0.25$, $p = 0.11$. A significant interaction effect ($F(6, 108) = 2.96$, $p < 0.01$) showed an increase in motivation scores by the lower scoring group across time and a decrease in motivation scores by the higher scoring group. No significant effect of time was found. Infants with higher AIMS scores had higher motivation to move scores than infants who scored lower on the AIMS. The t-test shows that strongly-motivated infants had earlier onset for all motor milestones (sitting, pulling-to-stand, hands-and-knees, crawling and cruising) than weakly-motivated infants ($t(13) = 2.39, 2.98, 2.25, 2.50$ $p < 0.05$). Infants' MTM score was positively correlated with the AIMS percentile at the same and subsequent sessions (Pearson correlations ranging from $r = 0.36$ to 0.69; with only $r = 0.36$ ns ($p = 0.06$).

Environmental factors					
Majnemer (2006, Canada)	n = 155 GA > 38 weeks *M* GA and *M* BW not reported	**Motor:** AIMS, PDMS, Battelle Developmental Inventory Age equivalent (mon) **Age:** 4 or 6 months, 15 months	**Factor:** sleep position **Measurement:** parental diary 3 consecutive days / 24 hours every 5 minutes **Age:** 4 or 6 months	Sex* Parental education * Parental age* Parity* Weight at assessment* Age at testing*	Linear regression showed there were no significant differences between sleep position on AIMS total score and PDMS score at 4 months. At 6 months of age, infants sleeping prone had significantly better motor scores on the AIMS total raw scores (p = 0.02) and PDMS (p = 0.03). At 15 months, no significant differences in PDMS score and Battelle Developmental Inventory Age equivalent scores. Linear regression models at 4 months shows that the AIMS prone raw score (r^2 = 0.27, p = 0.0001) and the total raw score were predicted by sleep position (prone versus supine) (r^2 = 0.17, p = 0.0001), when adjusting for confounders. When adjusting for confounders on linear regression models, sleep position consistently predicted AIMS motor scores and Peabody gross motor quotient, accounting for 22% to 31% of the variance. Univariate analyses indicated that the Battelle gross motor subscale score was significantly associated (p = 0.05) with sleep position, which was further demonstrated on simple linear regression analysis (r^2 = 0.8, p = 0.048). At 15 months of age, prone sleepers attained motor milestones significantly earlier: walking upstairs (p = 0.04) and walking (p = 0.05).

Supplementary Table S5.1 continues on next page.

Supplementary Table S5.1 ***Continued***

Author (year, country)	Number of participants and participant characteristics	Motor measures and ages at measurement	Factor measures and ages at measurement	Confounders (*in the final model)	Statistical analysis and results
Davis (1998, USA)	*n* = 351 *M* BW = 3490 g (*SD* = 41) *M* GA not reported	**Motor:** MM 9 motor milestones **Age:** 0–18 months	**Factor:** sleep position **Measurement:** position recorded by parents: prone and supine **Age measurement:** 2–6 months	BW* Sex* Maternal education* Ethnicity* Number of siblings*	Linear regression analysis shows that prone sleepers acquire motor milestones significantly earlier for: rolling prone to supine ($p < 0.002$), sitting unsupported ($p = 0.003$), creeping ($p = 0.0002$), crawling ($p = 0.003$) and pulling to stand ($p = 0.001$). Walking alone was not associated with prone sleeping ($p = 0.4$). Increased prone playtime was associated with tripod sitting, sitting alone, crawling and pulling to stand ($p < 0.05$). After controlling for maternal education, ethnicity, sex, BW and number of siblings, only pulling to stand remained significant ($p < 0.01$).
Lung (2011, Taiwan)	*n* = 1630 Birth cohort with 7.1% infants with chronic illness included *M* GA and *M* BW not reported	**Motor:** TBSC **Age:** 6, 18, 36 months	**Factor:** sleep position **Measurement:** interview at home **Age:** 6 months	Maternal education* Paternal education* Acute hospital admissions* Chronic illness*	At 6 months, structural equation model shows that infants sleeping supine had slower GMD ($b^* = -0.11$, $p < 0.001$). Supine sleeping position did not affect infant development at 18 and 36 months. Other factors were associated with infant GMD at 6 months: acute hospital admission ($b^* = -0.07$), chronic illness ($b^* = -0.05$) and paternal education ($b^* = 0.06$). At 18 and 36 months, maternal education ($b^* = 0.11$ and $b^* = 0.07$) and chronic illness ($b^* = -0.13$ and $b^* = -0.05$) were also associated.

Ratliff (2001, USA)	*n* = 205 GA < 34 weeks (range = 29.33–29.65) BW < 1750 g (range = 174–1257) *M* GA and *M* BW not reported	**Motor:** BSID 2nd edition **Age:** 4, 13 months corrected age	**Factor:** sleep position **Measurement:** question on infants' usual sleeping position **Age:** 4, 13 months corrected age	Maternal education* Ethnicity*, Days hospitalized*, Methyxanthine use*, Marital status* Head circumference* Other maternal and infant characteristics were potential confounders, but were excluded from analysis due to *p* > 0.2	Multiple linear regression analyses show that the PDI scores of PT infants at 4 and 13 months corrected age did not differ significantly between prone sleepers and supine or side sleepers in both adjusted and unadjusted analyses (4 months: p = 0.7371; 13 months: p = 0.1454). Individual items of the BSID show that supine sleepers were less likely than prone sleepers to receive credit for: maintaining head at 45° and 90° (p = 0.021) and lowering the head with control (p = 0.001).
Jardi (2017, Spain)	*n* = 154 GA ≥ 37 weeks BW ≥ 2500 g	**Motor:** BSID 2nd Edition **Age:** 6, 12 months	**Factor:** BF (exclusive BF, mixed feeding and total time BF) **Measurement:** 24-hour food diary and questionnaires **Age:** at birth, 1, 4, 6, 12 months	BW* GA* Sex* Maternal education Maternal age* Maternal SES* Head circumference at birth, 6 and at 12 months* Height at birth, 6 and at 12 months* Iron status at 6 and 12 months* Infant haemoglobin at 6 and 12 months* BMI at 6 and 12 months*	Multiple linear regression showed in the adjusted model, that exclusive BF during the first 4 months increased the PDI by 7.712 points (p = 0.019), while mixed feeding increased it by 6.393 points (p = 0.039) at 6 months. Higher GA and higher BMI increased the PDI scores (p = 0.005 and p = 0.024 respectively) At 12 months, the adjusted model showed that exclusive BF during the first 4 months increased the PDI by 7.223 points (p = 0.033), while mixed feeding did not significantly increase the PDI (b^* = 4.620; p = 0.160). Higher iron status at 6 months increased the PDI scores (p = 0.015).

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Supplementary Table S5.1 ***Continued***

Author (year, country)	Number of participants and participant characteristics	Motor measures and ages at measurement	Factor measures and ages at measurement	Confounders (*in the final model)	Statistical analysis and results
Michels (2017, USA)	*n* = 4270 Groups: PT = 17% FT = 83% *M* GA and *M* BW not reported	**Motor:** MM **Age:** 4, 8, 12, 18, 24 months	**Factor:** BF (exclusive BF, mixed feeding) **Measurement:** parent report **Age:** 4 months	Maternal factors Ethnicity* Education* Age* BMI* PPD* Paternal factors Education* Age* Infant characteristics Sex* Plurality* Rapid weight gain until 4 months postpartum* ASQ pass/failure at 4 months* postpartum* Conception via fertility treatment*	Accelerated Failure Time models reveal that feeding differences at 4 months do not greatly affect the timing of gross motor milestone achievement. After adjustment for confounders, infants who were fed solids in addition to breastfeeding achieved standing faster than infants exclusively breastfed at 4 months (AF: 0.93; 95% CI [0.87, 0.99]). After controlling for multiple testing, these differences were no longer significant. No differences were found for PT and FT infants.
Morris (1999, Brazil)	*n* = 262 Groups: LBW (1500–2499 g) *n* = 131 GA ≥ 37 weeks *M* LBW = 2338 g (*SD* = 152) HBW (3000–3499 g) *n* = 131 GA ≥ 37 weeks *M* HBW = 3210 g (*SD* = 142)	**Motor:** BSID **Age:** 6, 12 months	**Factor:** BF intensity in first 4 weeks or 5–26 weeks **Measurement:** frequency of BF **Age:** at birth, 6 and 12 months	BW* SES* Diarrhoea morbidity*	Weak and non-significant correlations were observed between BF intensity in weeks 1–4 and 6 months. There was no association between BF intensity over weeks 5–26 and PDI scores at 6 and 12 months. Multiple linear regression models, adjusted for confounders, showed that BF frequency over the first 4 weeks of life was significantly associated with motor development at 6 months in both LBW and HBW infants (b^* = 0.23; 95% CI [0.00–0.45]; p = 0.047).

Oddy (2011, Australia)	n = 2868 All infants eligible *M* GA = 38.8 weeks (*SD* = 2.13) *M* BW not reported	**Motor:** IMQ **Age:** 24, 26, 36 months	**Factor:** BF duration **Measurement:** parental questionnaire **Age:** 0–12 months	GA* Sex* Maternal education* Maternal age* Maternal smoking in pregnancy* Biological father living with family* Total family income* Total amount of stressful life events during pregnancy* Apgar score infant at 5 minutes*	Overall, t-tests show no significant differences in GMD of infants who were breastfed < 4 months and > 4 months. In subsequent analysis separated by sex, boys receiving BF < 4 months did have an increased risk for one atypical score on GMD at one time point between 0 and 3 years (OR 2.03; 95% CI [1.17, 3.50]; p = 0.011).
Smith-Nielsen (2016, Denmark)	n = 83 Groups: PPD-group n = 53: *M* GA 40.2 weeks (*SD* = 1.3) *M* BW 3466 g (*SD* = 450) Control group n = 83 *M* GA = 40.6 weeks (*SD* = 1.2) *M* BW = 3583 g (*SD* = 526)	**Motor:** BSID-III **Age:** 4, 13 months	**Factor:** maternal PPD **Measurement:** EPDS **Age of Measurement:** 4, 13 months	Sex* Maternal co-morbid personality disorder*	Multivariate analyses of variance (MANOVA) showed no significant effects of PPD on motor scales at 4 and 13 months. Also, after adjustment for confounders, the effect remained non-significant (at 4 months p = 0.187; at 13 months p = 0.562).
Sutter-Dallay (2011, France)	n = 515 BW < 2500 g = < 1% *M* GA and *M* BW not reported	**Motor:** BSID-II **Age:** 3, 6, 12, 18, 24 months	**Factor:** maternal depression **Measurement:** EPDS **Age:** 6 weeks, 3, 6, 12, 18, 24 months	GA* Maternal education level* Maternal age* Mean income* Parity* EPDS score*	Multivariate regression models revealed no concurrent association between EPDS scores and infant motor scores over the follow up (b^* = 0.60; 95% CI [-0.40, 1.60]; p = 0.24). This association remained non-significant after adjustment for EPDS score at the time of infant assessment.

Supplementary Table S5.1 continues on next page.

Supplementary Table S5.1 ***Continued***

Author (year, country)	Number of participants and participant characteristics	Motor measures and ages at measurement	Factor measures and ages at measurement	Confounders (*in the final model)	Statistical analysis and results
Lung (2011, Taiwan)	n = 1693 All infants eligible *M* GA and *M* BW not reported	**Motor:** BSID **Age:** 6, 18, 36 months	**Factor:** maternal mental health **Measurement:** Interview, SF-36 **Age:** 6 months	Maternal education* Parental income* Family support*	Structural equation analysis showed that maternal mental health at 6 months was not significantly associated with GMD of infants at 6, 18 and 36 months. The study revealed the association of GMD with several other factors like family support, prenatal income, maternal education.
de Borba (2015, Brazil)	n = 40 Groups: Infants with adolescent mothers *M* GA = 37.3 (*SD* = 2.7) *M* BW = 2914 (*SD* = 734) Infants with adult mothers *M* GA = 38.7 (*SD* = 2.4) *M* BW = 3194 (*SD* = 539)	**Motor:** AIMS **Age:** 3 assessments with an interval of 2 months between 0–18 months	**Factor:** maternal age Adolescent: 15–19 years Adult: 25–39 years **Measurement:** questionnaire **Age:** maternal age at infant birth	No confounders considered	Generalized Estimated Equations showed that AIMS percentile (F(938.2) = 0.003, p = 0.874) and total AIMS score (F(38.2) = 0.085; p = 0.755) did not differ between infants of adolescent mothers and adult mothers. Infants of adolescent mothers had lower scores in the third evaluation in supine position (p = 0.046).
Lung (2009, Taiwan)	n = 17595 *M* GA and *M* BW not reported	**Motor:** TBCS **Age:** 6, 18 months	**Factor:** parental mental health **Measurement:** SF-36 **Age:** 6 months	Parental education* Parental age*	Multiple linear regression showed that parental mental health (paternal and maternal) was not significantly associated with children's 6-month development (Paternal b^* = -0.01, t = 1.04, p = 0.298; Maternal b^* = 0.01, t = 0.74, p = 0.458). At 18 months, only maternal mental health was predictive of infants' GMD (Maternal b^* = 0.017, p = 0.01). When the covariates of parental education and age of childbirth were added, the effect of maternal mental health decreased (b^* = 0.02, t = 2.12, p = 0.034).

Hernandez (2011, Spain)	n = 72 *M* GA = 39.8 weeks (*SD* = 1.32) *M* BW = 3277.7 g (*SD* = 456.23)	**Motor:** BSID **Age:** birth, 12 months	**Factor:** parental neonatal perceptions **Measurement:** NPI **Age:** 3 days, 3 months	GA* BW SES Father and mother neonatal perception scores* NBAS (endurance item)*	Using stepwise multiple regression models, more negative maternal neonatal perceptions (b^* = -0.325, p = 0.024) and a higher GA (b^* = 0.340, p = 0.018) predicted psychomotor development at 4 months and accounted for 21.8% of the variance. At 12 months, paternal neonatal perceptions (b^* = 0.383, p = 0.010), together with the NBAS endurance item (b^* = 0.339, p = 0.021) were significant in accounting for 17.2% variance of the psychomotor development.
Siegel (1999, USA)	n = 109 *M* GA and *M* BW not reported	**Motor:** BSID, MM **Age:** 6, 9 months (n = 34) 9, 12 months (n = 35) 12, 15 months (n = 40)	**Factor:** use of a baby walker **Measurement:** exposure baby walker from parent interview **Age:** 6 and 9, 9 and 12, 12 and 15 months	Parental education	A three-by-three between-subjects MANCOVA showed a significant effect of walker experience on infants' motor milestones in general (multivariate $F[6,154]$ = 4.81 p = < 0.0005). The univariate test showed that the use of a baby walker significantly affects the developmental onset of sitting, crawling and walking ($F[2,79]$ = 11.07, 4.97 and 4.25, p = 0.0005, p = 0.01 and p = 0.02), with a later onset of the motor milestones. A significant main effect of the use of a baby walker was observed for motor and mental scores considered together (multivariate $F[4,196]$ = 6.16 p < 0.0005). The univariate tests showed significant effects for motor development ($F[2,99]$ = 6.06, p < 0.03). Parental education was added as a covariate in the analyses.
Souza (2010, Brazil)	n = 30 Groups: FT = 86.2% PT = 13.8% *M* GA, *M* BW not reported	**Motor:** BSID-III **Age:** 12, 17 months	**Factor:** daycare attendance **Measurement:** full time daycare attendance **Age:** 0–17 months	No confounders considered	Descriptive statistics showed that 13% (n = 4) of the infants attending daycare full-time had suspected delays in GMD at 12 and 17 months, according to the reference means of the BSID. Of these four infants, one infant was PT with LBW.

Supplementary Table S5.1 continues on next page.

Supplementary Table S5.1 ***Continued***

Author (year, country)	Number of participants and participant characteristics	Motor measures and ages at measurement	Factor measures and ages at measurement	Confounders (*in the final model)	Statistical analysis and results
Tsuchiya (2012, Japan)	*n* = 742 GA = 39.0–39.2 weeks BW = 2948–2985 g Infants with pathology affecting motor function *n* = 5 *M* GA and *M* BW not reported	**Motor:** MSEL **Age:** 6, 10, 14 months	**Factor:** seasonal variation **Measurement:** month of birth **Age:** at birth	GA BW Sex SES Parental ages Parity	In a linear regression model (month of birth is transformed to a trigonometric form), the season of birth was significantly associated with GMD at 6 months ($F(2,736) = 21.71$, $p < 0.001$ and 10 months ($F(2,736) = 12.36$, $p < 0.001$. At 14 months, the season of birth was not significantly associated with gross motor score ($F(2,736) = 1.21$, $p = 0.30$). Infants born in Mar-Apr show a peak in GMD and those born in autumn (Sep-Oct) show the lowest GMD scores. The cyclic fluctuation of motor development according to month of birth disappears at 14 months of age.
Vierhaus (2011, Cameroon / Germany)	*n* = 345 Groups: Cameroonian infants *n* = 73 German infants *n* = 272 *M* GA and *M* BW not reported	**Motor:** BSID III **Age:** 3, 6, 9 months	**Factor:** cultural context **Measurement:** N/A **Age:** time of inclusion	No confounders considered	Univariate analysis of variance of the BSID outcomes, depending on cultural background (Cameroonian Nso versus Germans, between subject factor) and cultural background by age (3, 6, 9 months, within subject factor), showed large differences between the two cultural backgrounds ($F = 65.58$; df 1/251; $p < 0.001$; $\eta^2 = 0.207$) in favour of the Cameroonian infants at 3 months. These differences decrease over time and are almost non-existent at 9 months ($F = 23.63$; df 2/502; $p = <.001$; $\eta^2 = .086$). The largest deviance is related to GMD at 6 months due to items as sitting and standing being reached by Cameroonian infants much earlier than German ones. The sequence of BSID items differ between the groups.

Bjarnadottir (2019, Denmark)	*n* = 650 GA > 37 weeks BW > 2500 g	**Motor:** MM 13 predefined milestones **Age:** N/A	**Factor:** BF, predictors pregnancy and birth, home environment **Measurement:** interviews/ questionnaires **Age:** ongoing parental interviews from 1 week – 24 months	GA* BW Sex* SES Maternal age* Maternal education Paternity leave*	Principal Components Analysis was used to analyze motor milestone outcomes, grouping the milestones into 'late' and 'early with late in opposite directions'. Multivariate analysis showed that sex, GA and maternal age (M = 0.32, p = 0.05. b^* = -0.23, $p < 0.001$ and b^* = 0.05, p = 0.02 respectively) were significant predictors for the achievement of later milestones (crawling, walking and standing). Boys achieved these late milestones at an earlier age. For the early milestones, GA (b^* = -0.11, p = 0.01) and paternity leave (M = -0.28, p = 0.01) were significant predictors. Linear and logistic regression analysis revealed that motor milestone achievement from 1 to 24 months was not significantly related to BF duration (exclusive or total).
Flensborg (2017, Denmark)	*n* = 5601 *M* GA = 39.1 weeks (range = 27–46.5) *M* BW = 3250 g (range = 850–5450)	**Motor:** MM **Age:** N/A	**Factor:** GA, BW and other predictors **Measurement:** questionnaire, measurements **Age:** at birth and 12 months	Sex	Multiple linear regression analysis showed that most of explained variance (14.5%) in motor milestone attainment is due to GA (b^* = -0.15; $p < 0.001$) and BW (b^*= -0.16; $p \leq 0.001$), after adjustment for confounders. Other predictors (p-values ≤ 0.10 were considered significant) in the final model were: BF, paternal age, higher birth order, weight increase (all negative associations) and larger head (positive association).

Supplementary Table S5.1 continues on next page.

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Supplementary Table S5.1 ***Continued***

Author (year, country)	Number of participants and participant characteristics	Motor measures and ages at measurement	Factor measures and ages at measurement	Confounders (*in the final model)	Statistical analysis and results
Pereira (2016, Brazil)	n = 49 Groups: Preterm n = 12 (24.5%) Term n = 37 (75.5%) *M* GA = 38.20 weeks (range = 32–42 weeks *M* BW = 3156 g (range = 2200–3995 g	**Motor:** AIMS **Age:** 3 assessments from 2–12 months	**Factor:** home environment, maternal practices, cognition **Measurement:** DAIS, A-HEMD-IS, KIDI **Age:** between 2–12 months	GA BW Sex* Cognition* DAIS score* Family income* Mechanical ventilation*	Generalized Estimating Equations used for longitudinal analysis showed that the scores on motor development increased over time and strongly significant correlations were found between the motor outcomes at the three time points. Multivariate analysis revealed at assessment 1 that: family income (p = 0.011), score on cognition (p > 0.001), days of mechanical ventilation (p = 0.099) and being put in more stimulating and independent positions (p = 0.037) explained motor performance significantly (Adj R^2 = 0.876). At assessment 2, the multivariate model included cognition (p > 0.001) and family income (p = 0.003, Adj R^2 = 0.860); at the third assessment, only cognition (p > 0.001) remained in the model (Adj R^2 = 0.751). Variability in motor development is better explained by environment and parental knowledge and practice.

Supplementary Table S5.2 Confounders of the included studies

	Child confounders									Environmental confounders								
Author (year)	Age	Anthropometry	Birth order	BW	Ethnicity	GA	Plurality	Sex	Other child factors	Marital status	Maternal age	Maternal education	Number of siblings	Parity	Parental income/SES	Paternal age	Paternal education	Other environmental factors
Child factors																		
GA																		
Preterm																		
Yaari (2018)								X										
Term																		
Espel (2014)			–	–	X	–		–										
Postterm																		
Field (1978)																		
Birthweight																		
Low BW																		
Datar (2009)		X_1	X	–	X	X			X_2	X		X			–		X	
Grantham McGregor (1998)															$–_s$			
Normal BW																		
Lung (2009)				X		X	X_{twin}	X				X			X			
Twin																		
Nan (2013)				–		X		X										
Brouwer (2006)				–		X												
Goetghebuer (2003)		X_3		X					X_4				X					
Wilson (1975)				X														

Supplementary Table S5.2 continues on next page.

Supplementary Table S5.2 ***Continued***

Author (year)	Child confounders									Environmental confounders								
	Age	Anthropometry	Birth order	BW	Ethnicity	GA	Plurality	Sex	Other child factors	Marital status	Maternal age	Maternal education	Number of siblings	Parity	Parental income/SES	Paternal age	Paternal education	Other environmental factors
Anthropometry																		
Weight, length, head circumference																		
Scharf (2016)				X		–		–										
Overweight																		
Slining (2010)	X	–						X	X_5									
Proportional larger head																		
Bartlett (1998)																		
Afro-American background																		
Capute (1985)								-							$-_s$			
Motivation to move																		
Atun-Einy (2013)																		
Environmental factors																		
Sleep position																		
Prone sleeping																		
Majnemer (2006)	X_{test}	X_6						X			X	X		X		X	X	
Davis (1998)				X	X			X				X	X					
Supine sleeping																		
Lung (2011)									$X_{7,8}$			X					X	$X_{7,8}$
Ratliff (2001)		X_9			X				X_{10}	X		X						X_{11}

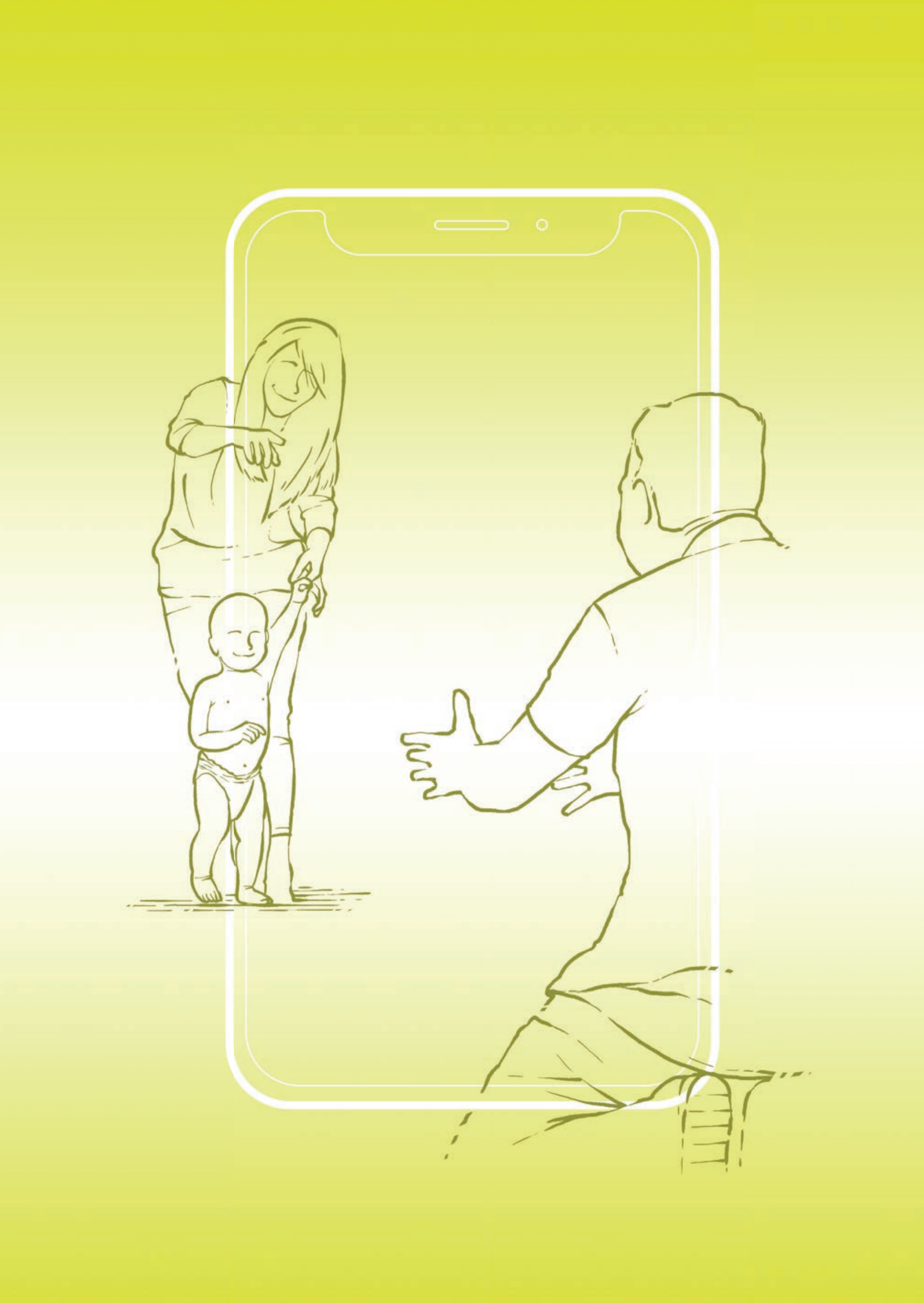
Author (year)	Child confounders									Environmental confounders								
	Age	Anthropometry	Birth order	BW	Ethnicity	GA	Plurality	Sex	Other child factors	Marital status	Maternal age	Maternal education	Number of siblings	Parity	Parental income/SES	Paternal age	Paternal education	Other environmental factors
Breastfeeding																		
Jardi (2017)		$X_{12,13,14,15,16}$		X		X		X			X	–			X_{s}			
Michels (2017)							X	X	$X_{20,21,22}$		X	X				X	X	$X_{17,18,19}$
Morris (1999)				X					X_{23}						X_{s}			
Oddy (2011)						X		X	X_{24}		X	X			X			$X_{25,26,27}$
Maternal depression																		
Smith-Nielsen (2016)								X										X_{28}
Sutter-Dallay (2011)						X					X	X		X	X			X_{29}
Maternal mental health																		
Lung (2011)												X			$X_{paternal}$			X_{30}
Adolescent mother																		
de Borba (2015)																		
Parental mental health																		
Lung (2009)											X	X				X	X	
Parental neonatal perception																		
Hernandez (2011)				–		X					–							$X_{31,32}$
Baby walker																		
Siegel (1999)												X					X	

Supplementary Table S5.2 continues on next page.

Supplementary Table S5.2 ***Continued***

Author (year)	Child confounders									Environmental confounders								
	Age	Anthropometry	Birth order	BW	Ethnicity	GA	Plurality	Sex	Other child factors	Marital status	Maternal age	Maternal education	Number of siblings	Parity	Parental income/SES	Paternal age	Paternal education	Other environmental factors
Daycare attendance																		
Souza (2010)																		
Season of birth																		
Tsuchiya (2012)				–		–		–			–			–	$-_s$	–		
Cameroonian vs German culture																		
Vierhaus (2011)																		
Multiple factors																		
Bjarnadottir (2019)				–		X		X			X							X_{33}
Flensborg (2017)								–										
Pereira (2016)			$X_{34,35}$	–		–		X										X_{36}

X = factor in final model; – = factor considered, but not in final model; -s = factor SES considered, but not in final model; 1= height; 2 = pregnancy / delivery risk factors; 3 = length; 4 = non-independence within twin pairs; 5 = age squared; 6 = weight at all assessments; 7 = acute hospital admission; 8 = chronic illness; 9 = head circumference; 10 = days hospitalized; 11 = methyxanthine use; 12 = infant iron status; 13 = infant hemoglobin level; 14 = head circumference at birth, 6 and 12 months; 15 = height at birth, 6 and at 12 months; 16 = BMI at 6 and 12 months; 17 = maternal BMI; 18 = maternal postpartum depression; 19 = maternal ethnicity; 20 = rapid weight gain until 4 months post-partum; 21 = ASQ pass/failure at 4 months; 22 = conception via fertility treatment; 23 = diarrhea morbidity; 24 = APGAR score infant at 5 min; 25 = total amount of stressful life events during pregnancy; 26 = biological father living with family; 27 = maternal smoking; 28 = maternal co-morbid personality disorder; 29 = EPDS score mother; 30 = family support; 31 = parental neonatal perceptions; 32 = NBAS (Neonatal Behavioral Assessment Scale) endurance item; 33 = paternity leave; 34 = cognition; 35 = Mechanical ventilation; 36 = DAIS score (Daily Activities of Infants Scale).



Chapter 6

Dutch parents and their beliefs on infants' gross motor development

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ABSTRACT

Objective: Parental beliefs (PBs) have an impact on motor development because they are the starting point from which parents act and make choices in raising their baby. This study explored changes in PBs about motor development of parents with term-born infants. The impact of infant birth order and motor trajectory on change was examined.

Design: The Parental Beliefs on Motor Development questionnaire (PB-MD) comprises five subscales reflecting a belief: *Stimulation*, *Natural Development*, *Advice*, *Own Pace*, and *Order* and were completed by parents at infant ages of 3.5 and 15.5 months. Infants were divided in three groups according to their individual motor trajectory, based on six motor assessments. A repeated-measures ANOVA was applied to explore changes in PBs, controlled for important covariates.

Results: Parents (N = 78) became significantly less attracted to the belief that active *Stimulation* of infant motor development was needed. An overall higher score on the subscale *Natural Development* was associated with infants in the more delayed motor developmental trajectory group. Furthermore, first-time parents were more drawn to the need for active stimulation of motor development of their infant than were experienced parents.

Discussion: PBs about motor development remain fairly stable between 3.5 and 15.5 months. The link between PBs, caregiving practices, and infant motor development needs further elucidation. Future research should include parents of infants at risk and parents from diverse cultural backgrounds and educational levels. PBs are a potentially modifiable factor to be addressed in new interventions that aim to optimize motor development.

INTRODUCTION

Infancy represents a period in life when parental caregiving is most intense and may therefore exert significant influence on infant development, especially motor development.[1] In everyday life with their baby, parents have many ideas about development and parenting that shape their interactions with their baby and the physical and social environment of the baby. These parental beliefs (PBs) are thought to have a profound effect on parental caregiving and hence on infant development. PBs can be defined as the reflection of ideas, thoughts, knowledge, and values that parents hold about children's development and socialization, parenting, and family life.[1,2] The pathways of influence of PBs on infant development through parental daily practices and habits are described in the 'developmental niche', a culturally originated framework.[2] According to this framework, three mutually interacting subsystems ultimately influence infant (motor) development: 1) the physical and social environment; 2) habits and customs in caregivers' practices; and 3) caregivers' psychology or PBs.[3,4]

PBs, especially those shared within a cultural community, have a great impact on physical and social environments and on habits and customs in caregiving practices. The choices parents daily make about caregiving practices and shaping the environment are mostly implicit, following cultural-specific patterns which are reflected in PBs.[5] In this way, culture shapes all three subsystems of the developmental niche model.

The third subsystem in the model, PBs, has not been the subject of many studies, especially those concerning infant gross motor development. Pereira and colleagues[6] examined the affordances in the home environment, maternal knowledge (with the Knowledge on Infant Development Inventory), and practices bearing on motor development (with the Daily Activities Scales of Infants), for 49 Brazilian infants between 2 and 12 months of age. They reported that, while maternal knowledge did not change significantly over time, maternal practices did. A regression model, with the variables infant cognition, gender, mechanical ventilation in the neonatal period, family income, and maternal practices, explained 88% of the variance in motor development in the first of the three assessments carried out over four months. Furthermore, significant relationships were found between maternal practices regarding infants' placement positions and their motor scores.[6]

A retrospective Brazilian study by Gomes et al.[7] investigated PBs about practices that stimulate motor development and how important parents perceive these practices to be. This study showed that most of the caregiving practices at the infant's age of

0–6 months were based on beliefs. In nine activities, the practices and beliefs did not converge, indicating the complexity involved in the formation of parental beliefs.[7]

In addition to the studies described above, several Dutch studies focused on PBs about child-rearing in general[5,8] and specifically about motor development.[9,10]

First, Harkness and colleagues[5] explored PBs and caregiving practices relating to arousal regulation of infants in five countries (Italy, Korea, Spain, USA, and the Netherlands). Fifteen Dutch families with a two-month-old baby were interviewed. The results demonstrated that the Dutch have a distinct cultural model of parenting that focuses on regularity of routines and a strong commitment to rest and sleep to stimulate the self-regulation of the baby. Besides, Dutch parents showed a more distant style of caretaking in which the baby's daily routine is mainly outlined in: spending time in a baby carriage, playpen, or bouncing chair.[5] These findings were confirmed in a recent qualitative study by van Schaik and colleagues[8] who found distinct differences in the ways Dutch mothers (n = 33) versus USA mothers (n = 41) approached the idea of getting the young baby into a schedule. During interviews, conducted when their baby was two or six months old, Dutch mothers discussed regularity in the day and night schedules significantly more frequently than did mothers from the USA. Also, daily schedules, detailed in diaries, revealed greater regularity for the Dutch babies.[8]

Second, with regard to PBs about motor development, the parental beliefs on motor development questionnaire (PB-MD) has been developed and cross-culturally validated in Israel and the Netherlands.[11] This was developed to objectify the PBs on motor development of parents with infants aged 1–8 months. A follow-up study was carried out to examine cultural differences in PBs between Israel and the Netherlands.[10] In this, the strongest predictor of PBs on motor development was cultural background. Parental factors (socio-economic status (SES), education, age) and infant factors (gender, birth weight) showed weaker relations with PBs. Dutch parents attributed less importance than did Israeli parents to 'stimulation' of motor development, 'advice' regarding motor development, and following motor development in the 'correct order'. Dutch parents agreed more with questions that expressed thoughts that children should follow their 'own pace'. Furthermore, cross-sectional data demonstrated that, compared to parents of younger infants, parents of older infants attributed less importance to stimulation and seeking advice and more to children's following their own developmental pace. Besides culture and age of the child, the study identified several other variables, such as gender, birth weight, parental education, and SES, and having seen a pediatric physical therapist (PPT). PB scores about *Natural Development* were higher for boys, and a higher birth weight predicted higher scores in the beliefs of parents on *Stimulation*. Parents with a higher SES were more attracted to seeking *Advice* and less to *Natural Development*. Finally, parents who visited a PPT were more drawn to the belief of seeking *Advice* and less to the idea that a child should follow its *Own Pace* in development.[10]

Where the study by van Schaik and colleagues[10] focused on the differences in PBs between Dutch and Israeli parents, the study by Oudgenoeg-Paz et al.[9] aimed to examine cross-cultural differences in actual practices to identify the pathways through which PBs influence infants' motor skills. Overall, the strongest relationship found between beliefs and practices was about the use of the prone position. Parents with stronger beliefs on *Stimulation* were found to apply more practices favoring the prone position and this was linked to more advanced infant motor skills in that position.[9,10]

To summarize, the above-mentioned studies indicate a link between culturally originated PBs and motor development in infancy. The findings suggest that parental care practices in physical and social contexts are important mediators in the relationship between PBs and how infants develop.

In the Netherlands, several recent studies revealed a delay in infant gross motor development compared to North American infants.[12–14] The average age at which Dutch babies walk independently is about two months behind their Canadian peers.[13] The origin of this finding may lie in cross-cultural differences in child rearing practices between these two Western societies. Given the evidence about the rest and regularity framework of Dutch parents,[5,8] further investigation of the PBs of Dutch parents about motor development may be very relevant.

To date, not much is known about the stability of PBs during a child's motor development. Because these are partially built on experiences and knowledge, PBs about motor development are likely to be subject to change in the first year of the baby's life. During this time, parents follow its development closely, often comparing this to that of other babies and receiving information from health care professionals as to whether their baby is developing according to usual standards. Furthermore, if parents already have a child, their prior experiences are also likely to shape their PBs about the motor development of their later-born infant. Therefore, it is hypothesized that the PBs of first-time parents will change more than those of parents who have had a child before. Thus, including birth order as a between-subjects variable may expand insight into the mechanism of changes in PBs. Previous research has only included parents with a first-born child to gain insight into pre-existing beliefs.[9]

To the best of our knowledge, there has been no research on the relationship between infant motor development and parents' change in beliefs about this during the first year of life. We hypothesized that new parents may start out thinking they will have a major influence on the motor development of their baby. If in time it turns out that their baby's development lags behind others, this may lead to adjustments in the beliefs they held at the birth, though the nature of such adjustments may differ individually. Possible changes include the following: 1) parents may reduce the importance they first attributed to motor development; 2) parents may no longer believe that they can influence motor development; or 3) parents may feel insecure and seek advice from a professional. The nature of the change in beliefs may thus be dependent both on the initial beliefs and on the rate of motor development of the infant.

Objectives of the study and research questions

The present study aimed to expand knowledge about (changes in) the beliefs that Dutch parents of healthy term-born infants have about motor development, focusing on the change in PBs between infant ages of 3.5 (T1) and 15.5 months (T2) and on factors that might predict change. Participants included first-time and experienced parents. In a previous study, individual gross motor trajectories of the participating infants were followed with six measurements between the ages of 3.5 and 15.5 months. Cluster analysis identified three groups with similar gross motor trajectories: *early developers*, *gradual developers*, and *late bloomers*. These motor development trajectory (MDT) groups were used in the present study.[15]

The research questions are:

1. Do the beliefs of Dutch parents on the motor development of healthy term-born infants change from 3.5 months to 15.5 months following birth?
2. Is there a significant effect of MDT group on parental beliefs?
3. Is there a significant effect of birth order on parental beliefs?
4. Is there a significant interaction effect of time (age of infant) and motor developmental trajectory group on parental beliefs?
5. Is there a significant interaction effect of time and birth order on parental beliefs?

METHODS

Design and participants

This study had a prospective longitudinal design and was part of a research project that explored gross motor trajectories of term-born infants from T1 to T2 (Figure 6.1).[15] Parents of healthy term-born infants were recruited through open registration between May 2016 and April 2018, leading to a convenience sample. Recruitment took place through flyers at birth centers, day-care centers for children, well-baby clinics, and maternity care offices in the larger cities of the Netherlands. Communication channels on social media were also employed to recruit parents. Parents of infants born before 37 weeks of gestation or diagnosed with pathology were not eligible to enter the study. Only parents with sufficient understanding of the Dutch language to read the informed consent forms and instructions were included in the study. Also, both parents or legal representatives had to sign informed consent forms. Ethical approval for the study was obtained from the medical ethics committee of the University Medical Center in Utrecht, the Netherlands (METC number 16/366C).

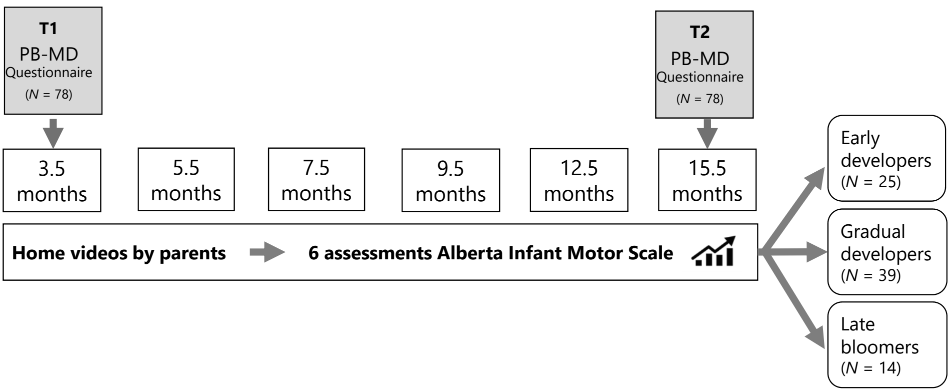

Figure 6.1 **Flow chart of the study design for parents (*n* = 78) and infants (*n* = 78).**

Instruments

Parental Beliefs on Motor Development questionnaire

To assess parental ideas and thoughts about the motor development of their baby, the Parental Beliefs on Motor Development questionnaire (PB-MD) was used.[11] Parents completed the PB-MD online when their infants were 3.5 and 15.5 months old. The PB-MD contains questions covering several themes regarding the thoughts parents have about parenting and the development of their baby. The key topics of the questionnaire revolve around the following questions: 1) How important is motor development for parents; 2) Do parents feel they have a role in advancing motor development; 3) Do parents think that stimulating motor development is necessary; and 4) Is expert advice on motor development needed? The themes were presented to parents through open questions, closed questions, and case descriptions. In the case descriptions and closed questions, parents rated their agreement on 6-point Likert scales (1 = totally disagree, 6 = totally agree). As a result of exploratory factor analysis, the outcomes of these questions are grouped into five main factors: 1) *Stimulation*, 2) *Natural Development*, 3) *Order*, 4) *Own Pace*, and 5) *Advice*. Face and convergent validity, test-retest reliability, and internal consistency had been found satisfactory for the Dutch population of parents with an infant between 1 and 8 months of age.[11]

Gross motor development

Gross motor development of infants was assessed with the Alberta Infant Motor Scale (AIMS) at 3.5, 5.5, 7.5, 9.5, 12.5, and 15.5 months, using home videos from parents. The AIMS home-video method was developed, successfully validated,[16] and perceived as feasible for parents, in longitudinal research.[17] Parents received checklists and instructional videos to help them capture the motor repertoire of their baby required for the AIMS assessment. They received digital reminders when it was time to make a new video, within a two-week window. Videos had to be uploaded to a secured streaming server which allowed trained PPTs to assess gross motor development with the AIMS.

The AIMS is a valid and reliable observation tool to assess the gross motor development of infants aged 0–19 months.[18,19] It comprises 58 items in four subscales: supine, prone, sitting, and standing. A total raw score can be converted into a percentile score and a *z*-score which are based on a Canadian norm population of 2202 infants.[20] In 2014, a re-evaluation showed that the original norm scores were currently still valid and usable.[21] Dutch norms for the AIMS for 1697 infants were established very recently.[14]

In summary, the study of Boonzaaijer and colleagues[15] showed infants with similar pathways of motor development, clustered into three groups: 1) *late bloomers*, 2) *gradual developers*, and 3) *early developers*. The *late bloomers* did not start accelerating before 9.5 months. Although this group caught up a lot in their motor development, the majority of these infants had not passed all the items of the AIMS at 15.5 months. The *gradual developers* showed a more even growth in motor scores over time. The trajectories of the *early developers* were characterized by rapid motor growth before the age of 9.5 months and the achievement of all test items before 15.5 months.

STATISTICAL ANALYSIS

Since the PB-MD questionnaire had not been applied previously to parents with 15.5-month-old babies, the outcomes were initially analyzed to evaluate the impact on the reliability of the questionnaire in this sample. Also, the longitudinal use of the PB-MD is new, making examination of the interrelationship of scores on the five subscales at both measurement points relevant. So, before analyzing the results of the PB-MD questionnaire, the internal consistency on an item and scale level was examined using Cronbach's alpha, and correlations between the scores of the five PB-MD subscales were calculated using Pearson Correlation Coefficients.

To answer the research questions, a 2 time (T1, T2) x 3 MDT group (*early developers*, *gradual developers*, *late bloomers*) x 2 birth order (1 = first-born, 2 = later-born) repeated-measures ANOVA was applied, with time as a within-subject variable, MDT group and birth order as between-subject variables, and the subscales of the PB-MD questionnaire as dependent variables. Previous research has shown that infant's *gender*, *parental age*, and *parental education*[9,10] affect PBs about motor development and these were therefore added as covariates. Because the population consisted of healthy, term-born infants, no effect of birth weight and gestational age was expected and therefore these variables were not included in the analysis.

Because the PB-MD does not allow for the conversion of subscale scores into a total score, interpretation of the multivariate results was not possible. Therefore, only the univariate outcomes of the repeated-measures ANOVA were interpreted. Bonferroni was applied to correct for multiple testing. The Statistical Package for Social Science 25 (SPSS) was used to analyze the data.

RESULTS

Descriptive statistics

Of 103 parents who participated in the research project,[15] 78 were included in this study, namely those who had completed both questionnaires. Reasons for dropout on the second questionnaire were not obtained. No significant differences in background characteristics were present between the total sample (n = 103) and the current sample (n = 78), except for the variable infant gender (p = 0.008). More parents with a boy dropped out, resulting in overrepresentation of parents with a girl (68%). Descriptive statistics were obtained to summarize the characteristics of parents and children (Table 6.1).

Table 6.1 Parent and infant characteristics

Infant characteristics		Mean (*SD*)/*n* (%)	Range	*n*
Gender	Female Male	53 (68) 25 (32)		78
Birth weight		3539g (424g)	[2780–4560g]	78
Gestational age		39.9 weeks (1.2)	[37–42 weeks]	78
Birth order	1st 2nd 3rd 4th	42 (54) 29 (37) 6 (8) 1 (2)		78
MDT group	Late bloomers *n* = 14	Gradual developers *n* = 39	Early developers *n* = 25	78
Gender (female)	7 (50)	28 (72)	18 (72)	
Birth order (first)	3 (21)	25 (64)	14 (56)	
Native language (not Dutch)	0	0	3	
Parent characteristics		**Mean (*SD*)/*n* (%)**		***N***
Parent completing PB-MD	Mother Father	77 (99) 1 (1)		78
Parental age	20–24 years 25–29 years 30–34 years 35–39 years 40–45 years	1 (1) 14 (18) 38 (49) 20 (26) 5 (6)		78
Parental education	No education Primary Lower secondary Higher secondary Tertiary	0 (0) 0 (0) 0 (0) 10 (13) 68 (87)		78
Parental professional classification	No profession Lower Secondary Higher Scientific	4 (5) 3 (4) 11 (14) 42 (54) 18 (23)		78
Native language	Dutch Other	75 (96) 3 (4)		78
Marital status	Married/living together Single Other	75 (96) 2 (3) 1 (1)		78

Reliability of the PB-MD in this population at T1 and T2

First, means and standard deviations of the subscales of the PB-MD were computed at T1 and T2. Cronbach's alpha values all exceeded 0.60, except for the subscale *Own Pace* (α = 0.55 at T1). In Table 6.2, the descriptive statistics of the PB-MD subscale variables and Cronbach's alpha values are displayed.

Table 6.2 Means, *SD* and internal consistency of the PB-MD subscales at T1 and T2

Subscales PB-MD	No. of items	T1 (*n* = 78) Mean (*SD*)	Cronbach's α	T2 (*n* = 78) Mean (*SD*)	Cronbach's α
Stimulation	6	2.39 (0.66)	0.65	2.25 (0.70)	0.71
Natural Development	3	2.91 (0.91)	0.62	3.34 (0.97)	0.60
Advice	3	2.71 (1.03)	0.69	2.72 (1.11)	0.68
Order	2	2.76 (1.36)	0.75	2.78 (1.38)	0.83
Own Pace	4	3.96 (0.86)	0.55	4.00 (0.93)	0.68

SD = standard deviation; T1 = 3.5 months; T2 = 15.5 months.

Correlations between the subscales of the PB-MD

Multiple subscale scores of the PB-MD were intercorrelated. The scores on each subscale at T1 and T2 all significantly correlated with each other, showing large effect sizes (r = 0.46–0.65, $p < 0.001$).[22] The subscale scores of *Natural Development* at T1 and T2 showed consistent positive associations with those for *Own Pace* at T1 (r = 0.51 and 0.31, $p < 0.001$, respectively) and T2 (r = 0.59 and 0.53, $p < 0.001$, respectively), with medium to large effect sizes. The subscale scores for *Stimulation* at T1 and T2 correlated positively with those for Advice and negatively with those for *Own Pace*, at both time points (Table 6.3).

Parental beliefs on motor development over time

6

Parental beliefs about stimulation

The covariate *parental education* significantly interacted with time ($F(1,77) = 10.74$, p = 0.002 partial $\eta^2 = 0.14$) on the subscale *Stimulation*. After controlling for the interaction of *parental education*, there was a significant main effect of time on *Stimulation* ($F(1,77)$ = 9.97, p = 0.002, partial η^2 = 0.88), showing that, between infants' ages of 3.5 and 15.5 months, parents become less attracted to the idea of active stimulation of motor development.

Table 6.3 Pearson Correlation Coefficient between PB-MD subscales at T1 and T2

	1.	2.	3.	4.	5.	6.	7.	8.	9.
1. PB-MD Stimulation at T1									
2. PB-MD Natural Development at T1	-0.14								
3. PB-MD Advice at T1	**0.28***	-0.16							
4. PB-MD Order at T1	-0.03	-0.09	**0.29****						
5. PB-MD Own Pace at T1	**-0.26***	**0.51****	**-0.43****	**-0.23***					
6. PB-MD Stimulation at T2	**0.62****	-0.19	**0.24***	0.02	**-0.28***				
7. PB-MD Natural Development at T2	-0.09	**0.46****	-0.03	-0.03	**0.31****	-0.21			
8. PB-MD Advice at T2	0.18	-0.17	**0.55****	**0.45****	-0.20	0.24*	-0.14		
9. PB-MD Order at T2	0.07	-0.03	**0.23***	**0.65****	**-0.23***	0.00	0.00	**0.56****	
10. PB-MD Own Pace at T2	-0.12	**0.59****	**-0.26***	-0.12	**0.57****	**-0.50****	**0.53****	**-0.29***	-0.02

T1 = 3.5 months; T2 = 15.5 months. * $p < 0.05$, ** $p < 0.001$.

Further, a significant main effect of birth order was found after controlling for the covariate *parental education* ($F(1,77) = 4.28$, $p = 0.002$, partial $\eta^2 = 0.06$), revealing that first-time parents were more drawn to the belief that stimulation of motor development is important than were experienced parents. No significant main effect of MDT group was found, nor was there an interaction effect between time and MDT group or between time and birth order.

Parental beliefs about Natural Development

The covariate *gender* was significantly related to *Natural Development* ($F(1,77) = 5.40$, $p < 0.05$, partial $\eta^2 = 0.07$) just as was *parental education* ($F(1,77) = 7.46$), $p < 0.01$, partial $\eta^2 = 0.10$). A significant main effect of MDT group on *Natural Development* was found ($F(2,76) = 3.68$, $p = 0.03$, partial $\eta^2 = 0.01$) after controlling for the effects of *gender* and *parental education*. A post hoc test revealed that parents of infants in the *late bloomers* group ($M = 3.55$) exhibited significantly higher scores on the belief of *Natural Development* compared to parents of the *early developers* ($M = 2.81$; mean difference = 0.75, $p = 0.049$). The *gradual developers* ($M = 3.26$) did not significantly differ from the *late bloomers* and the early *developers*.

Parental beliefs about Order, Own Pace, and Advice

For the subscales *Order*, *Own Pace*, and *Advice*, no significant main effects or interactions were found (Table 6.4).

Table 6.4 Final model with MDT groups and birth order

Main effects and interactions	PB-MD subscales	*F*(df)	*p*	Partial η^2
Time	Stimulation	9.97 (1,77)	0.002*	0.88
	Natural Development	0.14 (1,77)	0.91	0.05
	Advice	0.31 (1,77)	0.58	0.00
	Order	1.38 (1,77)	0.24	0.02
	Own Pace	1.11 (1,77)	0.30	0.02
Birth order	Stimulation	4.28 (1,77)	0.04*	0.06
	Natural Development	0.15 (1,77)	0.70	0.00
	Order	1.70 (1,77)	0.20	0.02
	Own Pace	0.25 (1,77)	0.62	0.00
	Advice	0.34 (1,77)	0.56	0.01
Time x Birth order	Stimulation	0.06 (1,77)	0.81	0.00
	Natural Development	2.17 (1,77)	0.15	0.03
	Advice	0.53 (1,77)	0.47	0.01
	Order	0.23 (1,77)	0.63	0.00
	Own Pace	0.38 (2,76)	0.54	0.01
MDT group	Stimulation	1.00 (2,76)	0.37	0.03
	Natural Development	3.68 (2,76)	0.03*	0.10
	Order	0.04 (2,76)	0.96	0.00
	Own Pace	1.88 (2,76)	0.16	0.05
	Advice	0.03 (2,76)	0.97	0.00
Time x MDT group	Stimulation	1.98 (2,76)	0.15	0.05
	Natural Development	2.05 (2,76)	0.14	0.06
	Advice	0.62 (2,76)	0.54	0.02
	Order	0.60 (2,76)	0.54	0.02
	Own Pace	1.25 (2,76)	0.29	0.04
MDT group x Birth order	Stimulation	1.40 (2,76)	0.25	0.04
	Natural Development	0.68 (2,76)	0.51	0.02
	Order	0.99 (2,76)	0.38	0.03
	Own Pace	0.24 (2,76)	0.79	0.01
	Advice	0.63 (2,76)	0.54	0.02
Time x MDT group x Birth order	Stimulation	1.57 (2,76)	0.22	0.04
	Natural Development	2.25 (2,76)	0.11	0.06
	Advice	0.20 (2,76)	0.82	0.01
	Order	1.56 (2,76)	0.22	0.04
	Own Pace	1.23 (2,76)	0.30	0.04

* Significant at $p < 0.05$.
1. Control variables: Gender (boy, girl), parental education (higher secondary and tertiary), parental age (20–24 years, 25–29 years, 30–34 years, 35–39 years, and 40–45 years).
2. Within-subject variables were added to the model simultaneously: MDT group (late bloomers, gradual developers, early developers); and birth order (first-born, later-born).

DISCUSSION

Parental beliefs are thought to have a profound impact on infant motor development and are an important factor to consider in understanding the variability that is present in early gross motor development. Studies on PBs about motor development are still scarce. Therefore, the primary goal of this study was to explore the change in beliefs about motor development of Dutch parents in the first year of life of their typically developing baby. An additional aim was to explore factors with a potential influence on any change in these beliefs.

This study demonstrated that PBs did indeed change between infants' ages of 3.5 and 15.5 months (T1 and T2), but only on one of the five subscales of the PB-MD, namely *Stimulation*. Dutch parents, during this period of 12 months, became less attracted to the belief that active stimulation of the infant's motor development was needed. Also, MDT groups had main effects on the subscale *Natural Development*, and birth order on the subscale *Stimulation*. No significant interaction effects were found for time and MDT group, and for time and birth order on the PB subscales *Natural Development*, *Order*, *Own Pace* and *Advice*, indicating stability of beliefs on gross motor development.

Parental beliefs about motor development

The mean subscale scores of the PB-MD at both T1 and T2 were lower than 3 on the 6-point Likert scale, except for the scores on subscale *Own Pace* (T1 and T2) and subscale *Natural Development* (T2). The scores of this sample correspond with former studies by van Schaik and colleagues[10] who also found lower scores (< 3) on the subscales *Stimulation*, *Advice*, and *Order* in 198 Dutch parents with an infant between 1 and 8 months. The variances found on the subscales *Stimulation*, *Natural Development*, and *Own Pace* also agreed with the results in the study of van Schaik et al.[10] The global pattern of scores on the subscales in the present study, namely lower ones on the subscales of Stimulation and Advice and higher on the subscales *Natural Development* and *Own Pace*, seem to agree with the framework of rest and regularity that was attributed to Dutch parenting in previous research.[5,8]

Overall, with change only present on one of five subscales, PBs on motor development seem to remain stable over time. This is in line with the findings of Winstanley et al.[30] who investigated the stability of maternal cognitions about child development at two time points, from birth to 5 months, in mothers (n = 105) of infants born term and preterm. They found that the parental cognitions were stable across time.

Parental beliefs about Stimulation

Comparison of the results of this study to others is challenging because PBs on motor development have not been evaluated longitudinally before. However, as time is also equivalent to the aging of the infant (from 3.5 months to 15.5 months), it is possible to relate the present outcomes to cross-sectional research concerning PBs on motor development and infants' age. In the study by van Schaik et al.,[10] including Dutch (n = 198) and Israeli parents (n = 206), infant's age was negatively related to beliefs about *Stimulation*. So, parents of younger infants more often thought that active stimulation of motor development was important than did parents of older infants. This is in agreement with the significant decrease over time in the scores on the subscale *Stimulation*, found in the present study. Van Schaik et al.[10] suggested that parents with an older baby adjusted their beliefs to a more realistic view of their impact on the infant's motor development. This suggestion might also apply to the outcomes in this longitudinal study. Another explanation for the decreasing scores on the subscale *Stimulation* might be that parents feel that a younger infant (about 0–8 months) is more dependent on the stimulation of motor activities initiated by parents, such as the provision of 'tummy time' when the infant is not yet able to roll over by itself. By the age of 15.5 months, most infants are capable of independent locomotion, which may lead parents to believe that active stimulation of the early motor milestones is less needed.

This study did not establish a relationship between the change in PBs about *Stimulation* and the MDT of the infant. This could be a result of the fact that only typically developing infants were included in the sample. Even though the *late bloomers* followed a more delayed gross motor trajectory, it remained a variation within the normal range and therefore the impact was perhaps too small to alter PBs on motor development. Future research into this question should include parents of infants with delayed motor trajectories.

In this study, no evidence was found to support birth order as a significant between-subject variable affecting the change in PBs on the subscale of *Stimulation*. The idea was that the change in PBs between T1 and T2 for first-time parents would be larger, in any direction and on any scale, than that of parents who had already experienced a first year of parenting. Nevertheless, there was a significant main effect of birth order on the *Stimulation* subscale, showing first-time parents to generally have higher scores. First-time parents were more drawn to the belief that motor development should actively be stimulated than were experienced parents. This implies that the beliefs of experienced parents are at least partly based on their experiences with their first-born child. Due to these earlier experiences, their ideas about motor development might be more realistic. However, this last suggestion is not supported by the outcome of the study of van Beek and colleagues[23] who evaluated the maternal expectations of Dutch and Italian mothers (*n* = 232) about the age of achievement of six motor milestones of their newborn term and preterm infants. The ages at which mothers expected the onset of the motor milestones did not differ between experienced and first-time mothers, not supporting the hypothesis that experienced parents have more realistic ideas about the onset of motor milestones than first-time parents. Lower scores on belief in active stimulation of motor development could also be a result of the fact that parents with a second or third child simply have less time than first-time parents. This makes active encouragement of the achievement of motor milestones more challenging.[24]

The covariate *parental education* showed a significant interaction with time on the subscale *Stimulation*. Additional plots revealed that the decrease in scores of the subscale *Stimulation* was largely driven by parents with higher secondary education whereas parents with a tertiary education remained stable in their beliefs. Several studies confirm that the level of parental education plays a role in PBs. In the study of van Schaik and colleagues[10] into the differences in PBs about motor development between Israeli and Dutch parents, parental education was a weak but significant predictor of beliefs. Harkness and colleagues[5] studied cultural models of self-regulation in parents of infants aged 0–2 months. In their qualitative study, this USA sample demonstrated that higher maternal education was associated with a greater emphasis on the stimulation of child development in general. This seems inconsistent with the current findings. However, because parents with only primary or lower secondary education did not appear in our sample, the nature of differences in Dutch PBs on motor development across all levels of education remains unknown at this moment.

Parental beliefs about Natural Development

The subscale *Natural Development* includes statements that active stimulation of motor development is not necessary and that development should take its own coarse. Over time, no significant change was found within this subscale. Interestingly, the main effect of the speed of infant motor development was on scores on the subscale *Natural Development*. Compared to parents with an infant in the *early developers* group, parents with an infant in the *late bloomers* group were more drawn to the belief that development occurs naturally and should not be interfered with. Longitudinal measurements of PBs allow for some interpretation of this finding about causality. The notion that associations between PBs and infant development are reciprocal and not just one way[25,26] complicates an unequivocal interpretation of this finding. Parents of the *late bloomers* might become more attracted over time to the belief that development proceeds naturally as a reaction to the repeated more negative feedback they receive on their baby's motor development between 3.5 and 15.5 months. Yet, in the present study, parents of the *late bloomers* did not show a change over time in this belief. At 3.5 months, when the first motor milestones of their baby were yet to be attained, these parents were already more drawn to the belief that development occurs naturally and should not be intervened with than were parents of the *early developers*. This indicates that, when the belief that motor development should take a natural course prevails, it may have some delaying influence on the gross motor development of term-born infants between 3.5 and 15.5 months. There is yet limited evidence linking PBs to developmental outcomes in infants. Further research is needed to explore this finding.

Parental beliefs about Own Pace

The items of the subscale *Own Pace* refer to the belief that parents should remain calm when the child is delayed, and the belief that a baby should not be forced to lie prone, especially when the baby starts crying in this position. Furthermore, the scores on this subscale remained stable across time which is not in line with the findings by van Schaik and colleagues,[10] who found a positive correlation between the infant's age and the scores on the subscale *Own Pace* in a sample of Dutch and Israeli parents.

The mean scores on this subscale were higher than those on all other subscales in this sample at both time points, indicating that parents are attracted to the belief that an infant should follow his/her own developmental pace. These relatively higher scores might also partly result from the information that parents receive from government well-baby clinics which are attended by about 92% of all Dutch children between 0 and 5 years.[27] In the Growth Guide issued by these clinics, a trusted source of information and advice, parents can read that it is normal for infants to follow their own pace in developing motor skills. Also, parents are advised not to compare their baby's developmental pace to others because every baby is unique.[28]

Parental beliefs about Order

There were no main effects or interactions with time on the subscale *Order*, nor were any main effects found of birth order and MDT group. This subscale refers to the belief that it is important that the infant's development follow a sequence in the attainment of motor milestones and does not miss one. In the present study, the mean scores on the subscale *Order* were higher than the Dutch sample in the study of van Schaik[10] but still < 3. Also, Dutch parents scored significantly lower on this subscale than did Israeli parents. Overall, SES and education were negatively associated with the subscale *Order*.[10]

To be able to appreciate this belief, parents should have some knowledge about the expected order of motor milestones. Although research on this topic among Dutch parents is lacking, the majority of parents are informed at well-baby clinics. Between 3.5 and 15.5 months, all newborns have six regular consultations scheduled, including a motor milestones screening. Furthermore, in the clinics' Growth Guide,[28] parents are informed explicitly not to worry about the order of development because some variation is considered normal (including some infants skipping a milestone). Overall, the lower mean scores and the fact that no interactions or main effects were found on this subscale might be a reflection of the low focus on the order of motor milestone achievement among Dutch parents.

Parental beliefs about Advice

On the *Advice* subscale, no main effects or interactions with time were found. In the cross-sectional study of van Schaik et al.,[10] the score on the subscale Advice was negatively associated with the age of the infant. In the present study, the score over time remained stable. Furthermore, the study of van Schaik et al. also found that having seen a PPT was positively related to the score on the subscale *Advice*. As our sample consisted of healthy term-born infants of whom only two had seen a PPT, it seems logical that in this study beliefs concerning *Advice* were not subject to change.

Limitations and strengths

The study outcomes are subject to some limitations. First, the present study administered the PB-MD at the infant ages of 3.5 and 15.5 months to first-time and experienced parents. The original sample on which the PB-MD was validated, consisted of 208 Dutch parents of infants aged between 1 and 8 months old and who were all first-born. Despite these differences in background variables, the internal consistency of the subscales remained acceptable, except for the outcome on the subscale *Own Pace* at 3.5 months. Second, participating parents were mostly well-educated, leading to a homogeneous sample. Another limitation was that, with one exception, only mothers completed both questionnaires. Even though, the composition of this study's sample is comparable to that with which the PB-MD questionnaire was validated,[11] this limits the generalizability of the study outcomes as the results will mostly reflect maternal beliefs on motor development.

A strength of this study was that, to the best of our knowledge, it was the first to examine the change in PBs about motor development over time. In doing this, insight was gained into how Dutch parents' beliefs on motor development behave over time. Also, this research was the first to relate motor developmental trajectories of infants from 3.5 to 15.5 months to changes in PBs on motor development, measured longitudinally. Even though the PB-MD questionnaire was originally developed and validated for cross-cultural research, this study demonstrated that the PB-MD is (partly) sensitive to change in parental beliefs in a mono-cultural sample and is also applicable to parents with infants older than 8 months.

Conclusion

This study shows that over 12 months, Dutch parents' beliefs on the motor development of typically developing infants remained rather stable, apart from those concerning stimulation. Across time, parents attributed less significance to the belief that active stimulation of motor development was needed. Birth order and the motor developmental trajectory group did not affect the change in PBs. Parents with a first-born child were more drawn to this belief than were experienced ones. The outcomes of this study roughly endorse the prevailing framework of rest and regularity among Dutch parents, as previously described.[5,8]

The finding that parents of late bloomers were more attracted to the belief of *Natural Development* in comparison to the parents of the *early developers* is an interesting result that calls for further exploration of the interlinkage between beliefs and infant motor development. Caregiving practices of parents in daily life with their babies are thought to be the link between PBs and infants' motor outcomes but were not addressed in the current study. They need to be added to future studies to gain insight into the way that context shapes infant motor development.[9]

The stability that was found in four subscales of the PB-MD may be consistent with existing evidence and endorses the theory that beliefs or cognitions are preferentially maintained.[29,30] However, the lack of observed change could also be linked to the limited diversity of the sample. Future research should include parents from diverse cultural and educational backgrounds and parents of infants at risk to further explore the changes in or stability of PBs about motor development.

PBs are considered a potentially modifiable factor that can be addressed in interventions aimed at improving infant motor development. From that perspective, future work could investigate whether PBs about motor development are indeed affected by interventions and whether changes in these subsequently positively influence the infant's motor trajectory.

In summary, this study provides a starting point for more in-depth research into PBs about motor developmental outcomes of both healthy infants and infants at risk. Understanding the role of PBs and caregiving practices in infant motor development can provide directions to new interventions that aim to improve early gross motor development.

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Author contributions

Marike Boonzaaijer: Conceptualization, Methodology, Investigation, Resources, Data Curation, Formal Analysis, Writing – Original Draft, Project administration. *Ora Oudgenoeg-Paz:* Conceptualization, Validation, Methodology, Formal Analysis, Writing - Review & Editing. *Imke Suir:* Conceptualization, Investigation, Writing - Review & Editing. *Jacqueline Nuysink:* Conceptualization, Methodology, Writing - Review & Editing. *Chiel Volman:* Conceptualization, Methodology, Formal Analysis, Writing - Review & Editing. *Marian Jongmans:* Conceptualization, Methodology, Writing - Review & Editing, Supervision.

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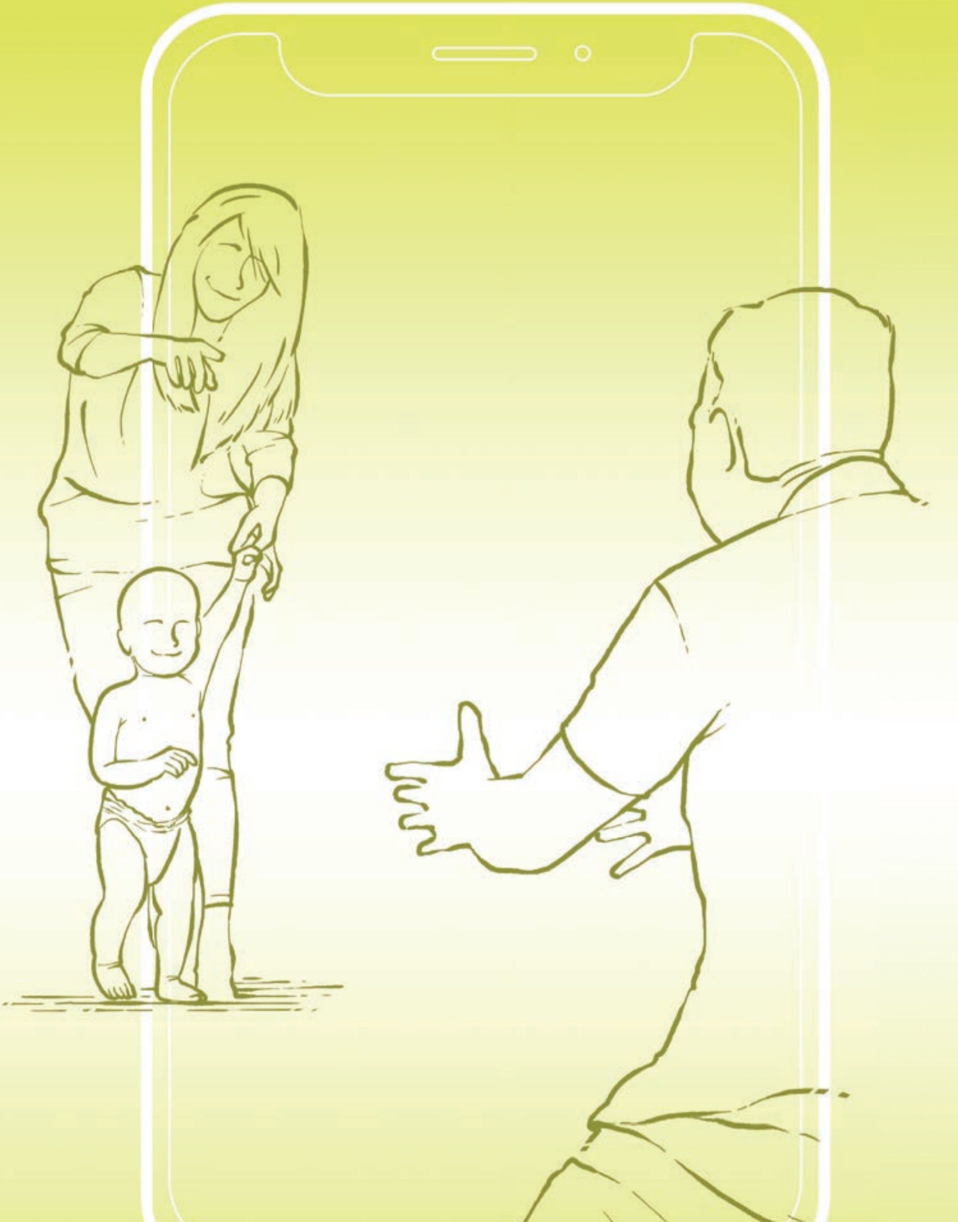

Chapter 7

Summary of findings and general discussion

The main objective of the studies reported on in this thesis was to gain insight into the early gross motor trajectories of typically developing (TD) infants. A better understanding of the variability in gross motor development, present within and between TD infants, will support clinicians to decide whether referral for early interventions is needed or not. Furthermore, a gross motor growth curve of TD infants during this early period can serve as a starting point for growth curves of infants at risk for delay and might support future studies that aim to evaluate the effects of interventions.

To enable longitudinal data collection a home video method, which revolved around the Alberta Infant Motor Scale (AIMS), was developed and validated. Subsequently, a pilot study was conducted to evaluate the expectations and experiences of parents who applied the AIMS home video method, using both questionnaires and interviews (Part I). After the validity and the feasibility of the home video method were determined, the method was utilized to gather data to model gross motor growth curves of term-born infants aged 3.5 to 15.5 months including six measurements with the AIMS (Part II).

To gain an overview of the existing evidence to date of both child and environmental factors associated with the variability in gross motor development, a systematic review with a unique focus on longitudinal studies was conducted. One of the factors of interest was parental beliefs on motor development. The change in these beliefs of parents at their infant's age between 3.5 to 15.5 months was examined, including the role of the infants' birth order and gross motor developmental trajectory (Part III).

In this final chapter of the thesis, the main findings of all studies are summarized followed by a more in-depth general discussion of selected themes, methodology, and a view on future research and clinical practice.

SUMMARY OF THE MAIN FINDINGS

Part I

In **Chapter 2**, we examined the concurrent validity of the AIMS home-video method. The outcomes of an AIMS assessment by a paediatric physical therapist (PPT) on-site were compared with those of one performed by another PPT from the home videos made by parents. The sample comprised 48 infants, aged from 1.5 to 19 months. Twelve PPTs interchanged roles, performing both live and video assessments with the AIMS. The mean difference in AIMS scores between the live- and video-observations was 0.46 items (*SD* ± 1.98), being not statistically significant ($p = 0.115$; 95% CI -0.116 to +1.033).

The intraclass correlation coefficient (ICC) agreement between the scores obtained by live and video observation was 0.99. The Standard Error of Measurement (SEM) was calculated as 1.41 and the Smallest Detectable Change (SDC) was 3.88 items. The inter- and intra-rater reliabilities of the AIMS home-video method were examined by three testers. The ICC agreement on the total raw scores between the three testers was high (ICC = 0.99, SEM = 0.92, SDC = 2.55 items). The intra-rater reliability of the video method also showed high agreement: the ICC on the total raw score was 0.99, the SEM was 0.96, and the SDC was 2.66 items. We concluded that assessment with the AIMS based on home video recordings was comparable to assessment by live observation and that the inter- and intra-rater reliabilities of the video assessments were excellent.

After determining the validity and reliability of the home-video method, we evaluated the feasibility of the home-video method for parents, using a prospective mixed-methods design (see **Chapter 3**). A longitudinal pilot study was set up with 45 parents participating, together with their TD infants aged between 0 and 19 months. In this pilot, parents were invited to video their infant five times in eight months and upload the videos to a secure web portal. After the videos were assessed with the AIMS by PPTs/researchers, parents received feedback on their infants' motor development. To gain insight into the expectations and experiences of parents during the pilot, we administered a questionnaire at the start and again when the study was finished (n = 34). In addition, eight parents agreed to an interview, which allowed us to gain more in-depth insight into their practical experiences and their feelings and thoughts about the home-video method. The results of the questionnaire showed that parents perceived the home-video method as not imposing any burden on the infant but found that it did require some effort from the parents. The interviews revealed that this parental effort was mainly in 1) finding the time to make the home videos, and 2) uploading them to the secure web portal. Parents reported experiencing joy in the one-to-one interaction with their baby while making the videos and they appreciated the professional feedback on the motor development of their baby. Furthermore, some parents reported increased awareness and insight into their baby's motor development. We concluded from this study that the AIMS home-video method was feasible for parents of TD children. The study revealed that most constraints were practical in nature and could be overcome in future applications.

Part II

In **Chapter 4**, the individual trajectories of gross motor development of 103 TD infants were modelled into a gross motor growth curve. A non-linear cubic function provided the optimal fit for the data ($F(1,571) = 89.68$, $p < 0.001$). Although none of the control variables had a significant effect on the growth curve, birth order showed a trend, indicating that infants who already had two or more older siblings tend to show slower motor development. Finally, based on a cluster analysis of the individual motor trajectories, three groups were identified with similar trajectories: 1) *early developers*, who showed fast motor growth from 3.5 months to 9.5 months of age, 2) *gradual developers*, who followed a more even growth between 3.5 and 15.5 months and 3) *late bloomers*, who did not start accelerating until after 9.5 months and showed a lot of catching up growth towards 15.5 months. Between the groups, significant differences in the trajectories were found when growth curves for each group were modelled. We concluded that the motor growth of TD infants based on the AIMS can be modelled by a non-linear function. None of the child and environmental factors had a significant effect on the position of the motor growth curve but a trend was found for the variable birth order. Infants with a higher birth order were less advanced in their motor development.

Part III

In **Chapter 5**, we presented an overview of the existing evidence from longitudinal studies on the associations between child and environmental factors and infants' gross motor development from birth to independent walking. In 36 studies, six child factors and 11 environmental factors were identified with either positive, negative, or no association with gross motor development. The longitudinal evidence revealed that associations with some factors were present at specific ages, but not at earlier or later ones. Strong evidence was found for a negative association between gross motor development and a low birth weight in healthy full-term and preterm born infants. Moderate evidence was found for a negative association for the factors overweight, shorter gestational age, and a positive association with prone sleeping and gross motor development. For the following associated factors, the existing evidence was conflicting: 1) twinning, 2) breastfeeding, and 3) supine sleeping. No evidence was found for an association between maternal postpartum depression and gross motor development of infants. For the other 10 factors, only one longitudinal study was available and therefore evidence was limited. We concluded from this systematic review that child factors have been studied extensively by means of longitudinal designs and show some clear associations with gross motor development. For most environmental factors, the available longitudinal evidence was still limited, and the measurements were less robust than those used to measure child factors.

In **Chapter 6**, we investigated a specific factor of interest concerning the gross motor development of infants but so far scarcely studied: parental beliefs (PBs) about the gross motor development of infants. In a prospective longitudinal study with 78 parents of TD infants, we explored the change in PBs on motor development and the associations of PBs with infants' birth orders and gross motor developmental trajectories from 3.5 to 15.5 months. The PBs were assessed with the Parental Beliefs on infant Motor Development questionnaire (PB-MD) when their babies were 3.5 months of age and again at 15.5 months. These parents also provided home videos that were used to model a gross motor growth curve for TD infants (**Chapter 4**). The infants were grouped into three clusters: 1) *early developers*, 2) *gradual developers* and 3) *late bloomers*. Only the scores on the subscale Stimulation (active stimulation of motor development is needed) decreased significantly over time. Neither birth order nor motor developmental trajectory group was associated with this change. A significant main effect of birth order on the subscale *Stimulation* revealed that first-time parents were more drawn to the belief about the need to stimulate motor development than were experienced parents. The motor development trajectory group showed a significant main effect on the subscale *Natural Development* (development occurs naturally and should not be interfered with) showing that parents of infants in the late bloomers' group had significantly higher scores on this scale than did parents with an infant in the early developers' group. We concluded that PBs on motor development of Dutch parents with a TD infant remained rather stable between 3.5 and 15.5 months. The link between PBs and infant motor development calls for further research.

GENERAL DISCUSSION

In what follows, two topics related to the main aim of the thesis are addressed: variability in gross motor development, and the role parents have in gross motor development. Reflections on the methodological aspects of the studies presented in this thesis are woven into the more theoretical parts of this general discussion. We end with recommendations for future research and clinical practice, and the overall conclusion of this thesis.

Variability in gross motor development of infants

Given the main objective of the thesis, i.e., to study gross motor development longitudinally, we chose dynamic systems theory (DST) as a framework for our studies. In this discussion, we will reflect on that choice and link the main features of the DST (as reported in **Chapter 1**) back to our research.

But to begin with, why is variability in motor development seen as such an important research topic? After the era of the maturation theories, describing and charting motor development mainly as a stable product of brain maturation,[1,2] the DST aims to understand how motor development happens by studying the impact of experiences and context.[3] Following the DST framework, it was recognized that variability in motor behaviour should no longer be interpreted as an inconsistency or error, but as an essential element that is needed in the adaptation to the requirements of the body or the environment.[4,5] Variability is present in both quantitative and qualitative aspects of gross motor development, with time (multiple assessments are needed to observe change) and context (determines the expression of motor behaviour) as the important denominators. The presence of variability can be interpreted both as a strong indicator of a healthy nervous system and as a sign of change, which is a key feature of development.[6] Thus, in the quest of finding the "holy grail" of early prediction of motor development, understanding variability is still very relevant.[7]

Variability and the AIMS

In this thesis, we chose the AIMS to measure gross motor development [8]. By applying the AIMS, our longitudinal study (**Chapter 4**) focused mainly on the interindividual variability in the rate of achieving new motor skills. By providing detailed descriptions of posture, support surface, and anti-gravity movements of each item, the AIMS requires a very diligent assessment of motor skills that goes beyond recording motor milestones or observing more crude motor items, as used in the Bayley Scales of Infant Development III-NL (BSIDIII-NL)[9] and the Van Wiechen Developmental assessment.[10] The AIMS does not measure the quality of movement specifically, in contrast to the General Movements Assessment (GMA) that observes the variability, fluency, and complexity of the movements as an important marker of healthy development[11] However, by using the AIMS also qualitative aspects are taken into account that are embedded in the descriptions of the items. Therefore, besides the statistical variability, the gross motor growth curves in Chapter 4, also reflect the underlying detailed information on both the quantity and quality of the observed motor skills.

In light of the DST framework, it is interesting to deliberate on whether the AIMS assessment allows for any variability in individual motor development? Although the items of the AIMS have a chronological order, and age-skills are displayed on a time line that was based on a Rasch analysis,[8] the assessor can capture a different sequence by marking a so-called 'window' on the observation form. This window, formed by two brackets, is placed around the items that represent the current motor repertoire of the infant. Because only the observed items within the window are scored, the assessment does give room for variability in the sequence of motor development. On the other hand, alternate modes of movement and locomotion, such as bottom shuffling, cannot be captured in an AIMS assessment.

Concluding, applying the AIMS in this thesis brought both strengths and limitations to the assessment. Despite the limitations in explicitly capturing qualitative variability, it provides a valid and reliable representation of individual developmental progress. Also, the observational character of the assessment was an important advantage because it improved the feasibility of the AIMS home-video method for parents.

Variability and the AIMS Home-Video Method

The AIMS home-video method enabled us to capture motor behaviour in a unique way: while the infant was at home, engaging in meaningful interactions with their parents. This contrasts with research performed in controlled situations at research institutes.[12,13] Since all motor behaviour, and the variability that goes with it, is linked inseparably to the context in which the movement takes place,[3,14] the use of home videos strengthened the ecological validity of the results.

We believe that by using the home-video method, constraints within the child, such as state of regulation during the assessment, were optimized. The naturalistic videos also grasped some of the direct environmental constraints such as the surface, toys, siblings, and parental interaction.

Following the DST, the need for ecological research, as being complementary to lab research, is recognized because of the impact of daily activities on motor behaviour.[15] In future research, measurements of full daily routines, both play and non-play activities, would be a valuable aid to arriving at an understanding of how motor development happens.[16] Natural contexts can reveal more about the opportunities an infant has to practise a motor skill and therefore have implications for observing the development of new motor skills in infancy.[17]

Variability: change over time

With the DST as their starting point, several authors have addressed the impact that timescales have on the view on variability.[5,18] There are so-called micro-developmental studies that use high-frequency measurements in just a few weeks to capture the variability of a transition in a sensitive manner,[3,19] in contrast to studies with a pre-post design that only determine change, without providing information about the process involved.[5] Our prospective longitudinal design was in-between, assessing motor development at six time points between the infants' ages of 3.5 and 15.5 months. The intervals between the assessments were two months (3.5–5.5–7.5–9.5 months) or, later, three months (9.5–12.5–15.5 months), because we aimed to follow infants until independent walking, without burdening participating parents with too many measurements.

Although not as fine-grained as micro-developmental studies, our study revealed nonlinearity and intra-individual variability in accelerations and decelerations in motor development between 3.5 and 15.5 months. This provided the basis for the different gross motor growth curves of the three groups of infants within the normal range: 1) *early developers*, 2) *gradual developers*, and 3) *late bloomers*. This knowledge can support clinicians in estimating whether observed motor behaviour is within the range of normal variation or whether referral to early intervention is indicated.

Variability and associated factors

In **Chapter 4**, both infant and environmental factors, associated with gross motor development, were acknowledged by controlling the gross motor growth curve for known background variables. None of these factors had an observable impact on the gross motor trajectories, most probably due to the homogeneous nature of the sample in terms of infant birth weight, gestational age, and parental education and background. This brings us to the principal limitation of our studies, the participation mainly of well-educated parents. Even though we put effort and attention into including parents and infants from a wide range of cultural and socio-economic backgrounds, we were only partly successful. We expect that the study design contributed to this result because it was demanding commitments by parents, in time (> 12 months of participation), in making and uploading home videos of their baby at six time points, and in completing questionnaires.

We realize that this has consequences for the generalizability of the results of our studies. Attention to this biasing phenomenon has been drawn by the publications about

WEIRD science (Western, Educated, Industrialized, Rich, and Democratic).[20,21] Evaluation of research samples showed that over 96% of studies in the field of behavioural science were performed on WEIRD samples, which represent only about 12% of the population, and that many of these studies' claims to universal validity are unjustified.[21]

Participation in research is not feasible or desirable for some parents, neither in the Netherlands nor across the world. To improve this, the shift to participatory research designs, based on equal partnership and involvement of parents, clinicians, and researchers is promising, just as the technological means for measuring are rapidly evolving worldwide. New online paradigms might break down geographical barriers and increase accessibility to research projects.

Summary: DST as the framework of this thesis

To the question of whether the DST as a framework has 'worked' in this thesis, we can conclude with a positive answer. Studying variability through the lens of the DST has enabled us to 'look' at the trajectories of early gross motor development and the variability that is present in these trajectories among healthy TD infants. We have endorsed findings about the non-linear nature of gross motor development in infancy in a design where infant and context were both acknowledged. In our study designs, we allowed for realistic complexity. However, not all features of the DST are represented in our studies, features such as 1) overall development, across the domains of motor development, cognition, social and/or language development, and 2) specific transitions in motor development and the mechanisms involved in these changes.

The DST is still and will remain a guiding framework for the study of (motor) development. However, it is important to keep translating this abstract and theoretical framework into applied studies with relevance for clinical practice.[22]

To gain a further understanding of variability in motor development, complex designs are needed to examine the interactions and contributions of both biological (i.e. body size and composition) and environmental factors (i.e. infant daily activities and caregiving practices) to variation in motor development.[23] For this purpose, robust and feasible measurements are needed that will stand in naturalistic environments. New assessment tools such as accelerometry seem to open doors to micro-developmental research in the infant's daily life and so bridge the gap with laboratory studies.

Parents and their role in motor development in infancy

By watching more than 600 home videos of moving babies, at home and in spontaneous interaction with their parent(s), the impact parents have on their baby's gross motor development has literally become very visible. Although the main focus of this thesis was on the gross motor development of infants, we were well aware from the start of the project that their parents would play a role in this development as being the adult(s) taking care of them for most of each day. In this thesis, we explored parents' role in 1) the assessment of their baby's motor development with the AIMS home-video method (**Chapter 2**), and 2) the motor development of their baby by studying their beliefs on motor development (**Chapter 6**). Parents also actively participated in the studies described in **Chapters 2** and **4** by providing the home videos of their moving baby that constituted the basis for the AIMS assessments.

Parents' role in the motor assessment of their infant

Parents as active participants in their baby's motor assessment is an important benefit of the AIMS home-video method that fits the shift in thinking about the role parents should have in early intervention, the importance of which has been emphasized by many authors.[24–27] Furthermore, several parents reported that the home-video method added to their knowledge and awareness about motor development (**Chapter 3**). Entrusting parents with the task of making the home videos at the start of the collaboration might emphasize the parents' role as equal partners in the intervention. Future research must show whether these positive findings among parents of TD infants also apply to parents of infants at risk of delayed or abnormal gross motor development.

Parents' role in their infants' motor development: parental beliefs

In this thesis, we took an ecological perspective to understand more about the role parents have in the gross motor development of their infant,[28,29] we are the first to examine change in parental beliefs (PBs) about motor development longitudinally (**Chapter 6**), using the PB-MD questionnaire.[30] We detected a significant change on only one of the five subscales of the PB-MD, indicating that PBs on motor development in this small Dutch sample of parents are not so prone to change, even during a period that parents witness huge changes in their baby's motor development. If we aim to address PBs as a potentially modifiable factor for interventions to optimize motor development, more research is needed into the change or stability of beliefs over time. As culture is the most important predictor of parental beliefs on motor development,[31] we need to include parents from diverse cultural backgrounds.

Beliefs and practices

Research from behavioural science shows the relationship between beliefs and practices to be very complex.[32] Not only psychological mechanisms[33] but many other factors, such as personality traits, social norms, and motivation,[34] contribute to behaviour outcomes, which makes it challenging to predict actual practices from beliefs.

To investigate the pathways between PBs on motor development, caregiving practices, and infant motor development requires more research. To avoid the interference of other factors that might diminish the visibility of any relationship, the beliefs, practices, and development must all be measured at the same level, in the same specific domain or aspect of motor development. For example, if we measure the PB in stimulation, this should be combined with data on the parental practice of providing tummy time, and the infant's skills when lying in the prone position.[35]

With regard to practice, Bornstein's words seem to be very relevant: 'If parents do not think they can influence the development of their infant, they will act accordingly'.[31, p3] Thus, for clinicians working with parents, it is crucial to know parents' thoughts and knowledge about the motor development of their babies. In the PB-MD questionnaire, this is addressed in two questions: 1) Do parents have a role in supporting their baby's motor development, and if so, what is their role?, and 2) Should parents do something with their baby and/or with the environment to support the baby's motor development in the first year of life? If so, what should they do?.[29] These questions might be a good starting point for a conversation between parents and clinicians about PBs on motor development.

RECOMMENDATIONS FOR CLINICAL PRACTICE AND FUTURE RESEARCH

Part I

Our findings on the validity, reliability, and feasibility of the AIMS home-video method with TD infants and their parents, provide a strong indication that it could also be applied in clinical practice and follow-up clinics as a valuable addition to the usual care provided by a PPT. The recent COVID pandemic unintentionally underlined the relevance and value of new digital means, such as the AIMS home-video method, when most PPTs were suddenly forced to work from a distance with parents who had requested help. No software applications are yet available on the market to securely exchange video material between parents and professionals, an absolute requirement for use of the home-video method in a way that meets health care privacy regulations.

As we studied the validity and feasibility of the home-video method with parents who had not requested help with their babies, more research is needed to see if the home-video method is also acceptable for parents who have an infant at risk or have doubts or concerns about the progress of their baby's gross motor development. Furthermore, in an additional pilot within the GoAPP project (**Go**divApp **A**pplied in **P**ediatric **P**rimary care), following the GODIVA project, we have found that personalized instructions would be of great value. Some parents preferred the instructional videos, while others preferred the checklists on paper or digital.

About 600 home videos of infant motor behaviour in the context of the home were collected during this research project but used only to assess the AIMS. These video data, scientifically seen as transparent and sustainable, have the potential to answer new research questions within and beyond the domain of motor development. Novel directions in this area are presented by Databrary, a large-scale video repository where developmental psychology researchers can reuse such data to address new questions.[36]

Part II & III

Can we apply the findings from our studies (**Chapters 4**, **5**, **6**) to clinical practice, given that we only studied healthy term-born infants? In clinical practice and research, early identification of infants at risk is an important goal. However, to be able to properly identify delayed or abnormal motor development, we need to know what normal motor development looks like.

An informed decision about (non-)referral or intervention can be supported by the results of our longitudinal study (**Chapter 4**) and systematic review (**Chapter 5**) that highlight the broad range present in infant motor development and emphasize the need to consider both infant and contextual factors. As shown in previous studies and confirmed here, developmental surveillance is essential, since gross motor development is a non-linear process.[37]

Besides, the care needs of TD infants and their parents should not be forgotten. At well-baby clinics and in PPT practices, parents of TD infants raise questions and concerns. These concerns are mostly founded either on expectations that are not met or on comparisons with the development of other infants. Supporting parents in their worries and providing assessment, advice or information are important roles that prevent over- or under-stimulation of development and (para)medical 'shopping.'

In research, prospective longitudinal studies are needed to continue to learn about changes and transitions in the gross motor development of infants and factors associated with these. To add to practical relevance, the focus of these studies should be on potentially modifiable factors in the environment with the idea of improving interventions. More robust and continuous measurements should be developed and deployed that transcend the outcomes (in both reliability and level of detail) of methods such as retrospective parental diaries.[38] With the emergence of big data technology and artificial intelligence, new opportunities have arrived to collect reliable and rich data continuously that can provide more detailed insights into the motor activities of infants and parental caregiving practices in daily life. Another promising tool for data collection is the Ecological Momentary Assessment (EMA). By sending multiple short text messages to parents' smartphone, prospective data can be collected without much effort (time and distance are no barriers), providing a true reflection of infant behaviour, experiences, and affordances in the environment at any specific moment.[38]

As described in Chapter 1, besides the variability in quantity, the quality of motor behaviour is also an important feature in the detection of early motor delay. However, at present the quality of movement is not easily objectified in an assessment because the outcome largely depends on the experience of the clinician and the state of the infant.[39] Accelerometry and automated video analysis have the potential to improve this. In 2002, Esther Thelen stated: '*It may be that our visions of what questions can be asked are limited by the means we have to answer them*'.[3, p76] In 2021, we can add that new technological possibilities have expanded so rapidly and beyond expectations that, before measuring everything, it is particularly important to formulate appropriate questions such as: What aspects of the quality of infant movement are thought to be most predictive for motor development?

CONCLUSION

Returning to the original aim of this thesis, the two questions we addressed were: 1) can we use home videos made by parents to facilitate longitudinal data collection on gross motor development? and 2) can we model a gross motor growth curve and study factors associated with gross motor development?

To the first question, we can answer wholeheartedly "Yes." The AIMS home-video method has turned out to be a successful research tool in longitudinal data collection. For practice, it promises benefits for infants, parents, and PPTs.

Concerning the second question, we have expanded knowledge about the interindividual variability of TD infants in the rate of developmental progress according to the AIMS. We have studied this over time, acknowledging infant and environmental characteristics. For one of those environmental factors, PBs on motor development, we have examined change over time. In future research, the mechanisms of the impacts of PBs and caregiving practices on infant motor development deserve attention and should be studied in diverse samples with robust measurements of daily activities. New possibilities, such as the home-video method, but also accelerometry, and automated video analysis, are promising research tools to further improve our understanding of gross motor development in infancy.

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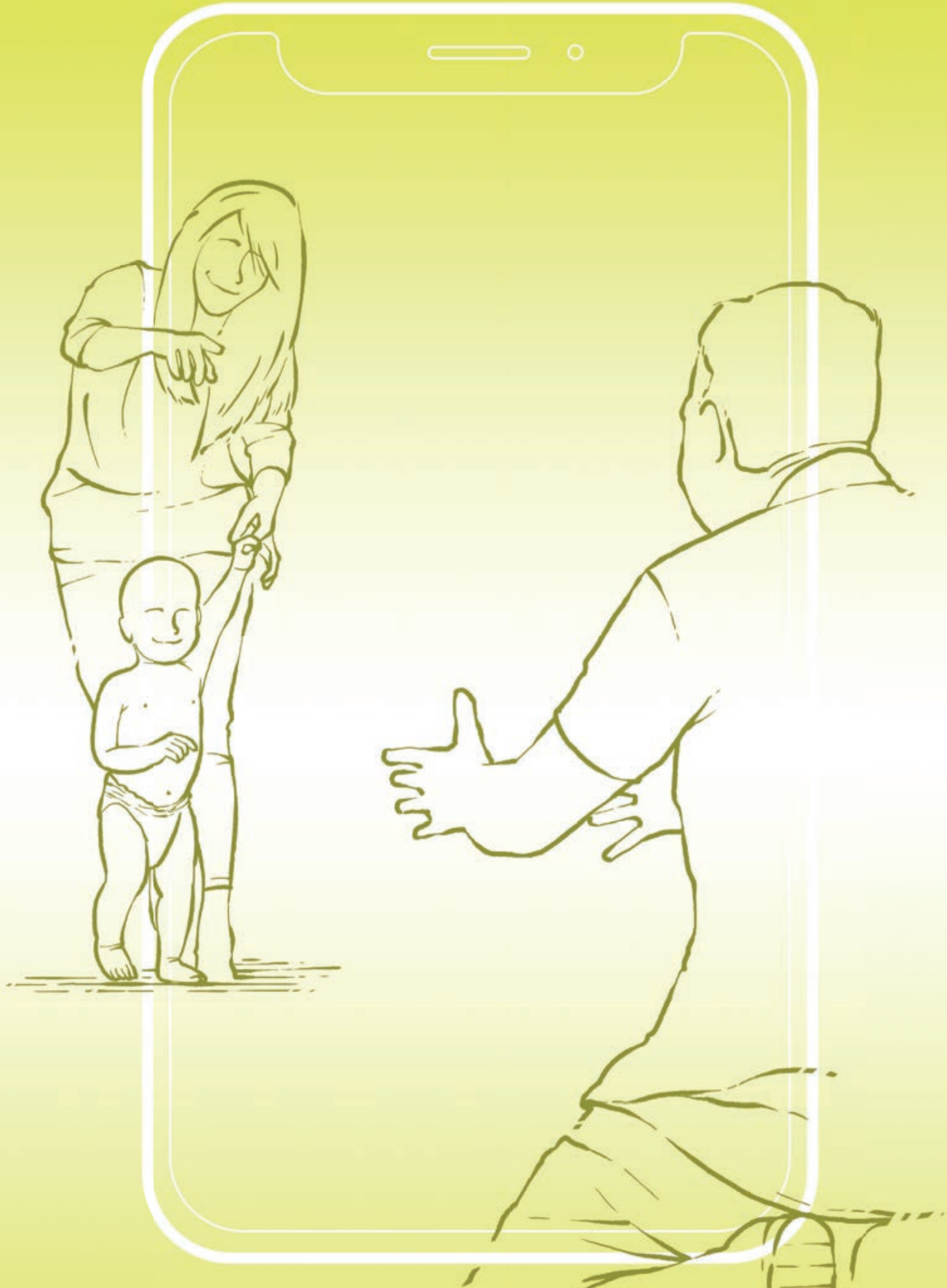

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SAMENVATTING

Achtergrond

Jaarlijks worden er in Nederland ongeveer 165.000 baby's geboren. Vanaf de geboorte wordt hun groei en ontwikkeling gemonitord op het consultatiebureau. De vroeg-motorische ontwikkeling is daarbij een belangrijke indicator die inzicht geeft in het functioneren van het centrale zenuwstelsel en de algehele gezondheid van het jonge kind. Omdat er een grote variatie zit in de ontwikkeling van de grove motoriek, is het accuraat vaststellen van een vertraagde of afwijkende motorische ontwikkeling een uitdaging. Dit heeft als gevolg dat er niet altijd tijdig wordt gestart met het behandelen van de kinderen die het meeste baat zouden hebben bij vroege interventie. Aan de andere kant worden er soms ook kinderen onnodig doorverwezen naar de kinderfysiotherapeut.

Om een duidelijker beeld te krijgen van de motorische ontwikkeling van kinderen is het belangrijk deze in de tijd te meten in plaats van op één moment. Dit herhaaldelijk observeren en testen kan belastend zijn voor zowel het kind als de ouders. Om deze belasting te verminderen is in 2014 het GODIVA-onderzoeksproject (Gross mOtor Development of Infants using home-Video registration with the Alberta Infant Motor Scale) gestart. In dit project is een methode ontwikkeld waarbij een motorische ontwikkelingstest, de Alberta Infant Motor Scale (AIMS), kan worden afgenomen door kinderfysiotherapeuten op basis van filmbeelden die thuis worden gemaakt door ouders.

Doel van het proefschrift

In de introductie (**Hoofdstuk 1**) wordt het doel van dit proefschrift als volgt beschreven: Inzicht verkrijgen in de interindividuele variabiliteit in de grof-motorische ontwikkeling van op tijd geboren kinderen door: 1) het modeleren van een grof-motorische groeicurve op basis van longitudinale metingen met de AIMS met behulp van de homevideomethode, en door 2) factoren te bestuderen die geassocieerd zijn met de grof-motorische ontwikkeling vanaf de geboorte tot zelfstandig lopen, waaronder de ideeën en overtuigingen van ouders, de Parental Beliefs (PBs).

Deel I

In **Hoofdstuk 2** onderzochten we de validiteit van de nieuw ontwikkelde homevideomethode. De uitkomsten van een AIMS-afname door een kinderfysiotherapeut ter plaatse werden vergeleken met de uitkomsten van een AIMS-afname op basis van de homevideo's gemaakt door de ouders en uitgevoerd door een andere kinderfysiotherapeut op een later tijdstip. De steekproef bestond uit 48 kinderen (1,5–19 maanden). Twaalf kinderfysiotherapeuten wisselden van rol in het uitvoeren van zowel de live- als de videobeoordelingen met de AIMS. De uitkomsten lieten zien dat het gemiddelde verschil in de AIMS-scores tussen de live-observaties en de video-observaties niet significant was. Ook werden er geen systematische verschillen gevonden. De intraclass correlatiecoëfficiënt tussen de scores van de live- en de video-observaties was hoog en de standaard meetfout lag binnen het vooraf vastgestelde criterium van twee items. Ook was zowel de intra- als de interbeoordelaarsbetrouwbaarheid van de AIMS homevideomethode goed. Wij concludeerden dat de beoordeling van de motoriek met de videomethode vergelijkbaar is met een AIMS-afname door middel van een live observatie.

In een longitudinale pilotstudie (**Hoofdstuk 3**), waaraan 45 ouders samen met hun kind (0–19 maanden, > 37 weken zwangerschapsduur) deelnamen, evalueerden wij vervolgens de toepasbaarheid van de homevideomethode voor ouders. Ouders werd gevraagd de motorische ontwikkeling van hun baby vijf keer in acht maanden tijd te filmen. Door middel van vragenlijsten bij aanvang en na afloop van het onderzoek (n = 34) en interviews (n = 8) zijn de verwachtingen en ervaringen van ouders onderzocht. Uit de resultaten van de vragenlijsten bleek dat ouders de homevideomethode als weinig belastend voor hun baby hadden ervaren, maar dat het wel enige inspanning van de ouders vroeg. Uit de interviews kwam naar voren dat dit vooral te maken had met 1) het vinden van het juiste moment om de video's te maken, en 2) het uploaden van de video's naar het beveiligde webportaal. Ouders waren positief over het plezier dat ze beleefden aan de één-op-één interactie met hun baby tijdens het maken van de video's en ze waardeerden de feedback over de motorische ontwikkeling van hun baby. Bovendien waren er ouders die rapporteerden dat de instructies hadden geleid tot een toegenomen bewustzijn van, en inzicht in de motorische ontwikkeling van hun baby. De meeste obstakels die ouders aangaven ten aanzien van de homevideomethode waren praktisch van aard. Wij concludeerden uit deze studie dat de AIMS homevideomethode goed uitvoerbaar en acceptabel is voor ouders van op tijd geboren kinderen.

Deel II

In Deel II van dit proefschrift is de AIMS homevideomethode ingezet als middel om de individuele grof-motorische trajecten van op tijd geboren kinderen van 3,5 tot 15,5 maand zichtbaar te maken. In **Hoofdstuk 4** zijn de trajecten van 103 kinderen gemodelleerd in een grof-motorische groeicurve. Een kubische functie paste het best bij de data. Hoewel geen van de controlevariabelen (geboortegewicht, zwangerschapsduur en geboortevolgorde van het kind en leeftijd, opleidingsniveau van moeder) een significant effect hadden op de groeicurve, liet de geboortevolgorde een trend zien, waarbij kinderen met twee of meer oudere broers of zussen vaker een langzamer motorisch traject lieten zien. Ten slotte werden op basis van een clusteranalyse van de individuele motorische trajecten drie groepen geïdentificeerd met verschillende trajecten: 1) vroege ontwikkelaars, die een snelle motorische groei vertoonden van 3,5 tot 9,5 maanden leeftijd, 2) geleidelijke ontwikkelaars, die een meer gelijkmatige groei volgden tussen 3,5 en 15,5 maanden en 3) laatbloeiers, die pas na 9,5 maanden begonnen te versnellen in hun motorische ontwikkeling en veel inhaalgroei vertoonden tot de leeftijd van 15,5 maanden. Wanneer de groeicurven per groep werden gemodelleerd bleken de verschillen tussen de groepen significant te zijn.

Deel III

De systematische review in **Hoofdstuk 5** geeft een overzicht van de bestaande evidentie uit longitudinale studies naar de associaties tussen de grof-motorische ontwikkeling van kinderen en kind- en omgevingsfactoren. De kwaliteit van de studies werd beoordeeld met de QUIPS (Quality in Prognostic Studies). Er werd sterk bewijs gevonden voor een negatieve associatie tussen een laag geboortegewicht en de grof-motorische ontwikkeling bij zowel op tijd geboren als prematuur geboren kinderen. Voor de factoren zwangerschapsduur, overgewicht, en het slapen in buikligging werd matig bewijs gevonden voor een associatie met de grof-motorische ontwikkeling. Voor de invloed van tweelingzwangerschap, borstvoeding en het slapen in rugligging was er tegenstrijdig bewijs aanwezig. Er werd geen bewijs gevonden voor een associatie tussen een postnatale depressie van moeder en de grof-motorische ontwikkeling van kinderen. Omdat er voor de overige 12 factoren maar één longitudinale studie beschikbaar was, blijft het bewijs voor deze factoren beperkt. Wij concludeerden uit deze systematische review dat er voor de kindfactoren (geboorte)gewicht en zwangerschapsduur matig tot sterk bewijs aanwezig is voor een associatie met de grof-motorische ontwikkeling. Voor de omgevingsfactoren waren niet alleen een beperkt aantal longitudinale studies beschikbaar, ook de gebruikte meetinstrumenten om deze factoren in kaart te brengen waren minder robuust.

In **Hoofdstuk 6** hebben we in een prospectieve studie met 78 ouders van op tijd geboren kinderen, de verandering in de ideeën en overtuigingen (Parental Beliefs) over motorische ontwikkeling onderzocht. De Parental Beliefs on Motor Development vragenlijst (PB-MD) werd afgenomen wanneer hun kind 3,5 maanden oud was en opnieuw met 15,5 maanden. Hieruit bleek dat alleen de scores op de subschaal Stimulatie ('*actieve stimulatie van de motorische ontwikkeling is nodig*') significant afnamen in de tijd. Deze verandering was niet geassocieerd met de geboortevolgorde of met de verschillende motorische trajectgroepen (vroege ontwikkelaars, geleidelijke ontwikkelaars en laat-bloeiers). Een significant hoofdeffect van de geboortevolgorde op de subschaal Stimulatie toonde aan dat ouders met een eerste kind zich meer aangetrokken voelden tot de overtuiging van het stimuleren van de motorische ontwikkeling in vergelijking met ervaren ouders. Een significant hoofdeffect van de motorische trajectgroepen op de subschaal Natuurlijke ontwikkeling ('*de motorische ontwikkeling verloopt natuurlijk en er moet niet worden ingegrepen*') liet zien dat ouders van een kind in de laatbloeiersgroep, significant hogere scores hadden op deze schaal in vergelijking met ouders met een kind in de groep van de vroege ontwikkelaars.

Wij concludeerden dat PBs over de motorische ontwikkeling van Nederlandse ouders met een op tijd geboren kind tussen leeftijd van 3,5 en 15,5 maanden tamelijk stabiel bleven. De geboortevolgorde en de motorische ontwikkeling van de kinderen waren niet van invloed op de verandering in de PBs maar zij hebben wel een relatie met de ideeën en overtuigingen die ouders hebben ten aanzien van de motorische ontwikkeling. In vervolgonderzoek moet dit nader onderzocht worden.

Hoofdstuk 7 bevat een overzicht van de uitkomsten van de studies en de discussie van dit proefschrift. In deze discussie kijken we terug op het gebruik van de dynamische systeemtheorie als basis voor de studies in dit proefschrift. Ook worden methodologische keuzes en beperkingen van de studies beschreven. Het hoofdstuk wordt afgesloten met aanbevelingen voor de praktijk en toekomstig onderzoek.

LIST OF ABBREVIATIONS

AIMS	Alberta Infant Motor Scale
PPT	Pediatric Physical Therapist/ Therapy
GMA	General Movements Assessment
MD	Motor Development
TD	Typically Developing
GMD	Gross Motor Development
GA	Gestational Age
BW	Birthweight
ABW	Adequate Birthweight
LBW	Low Birthweight
HBW	High Birthweight
MLBW	Medium Low Birthweight
VLBW	Very Low Birthweight
NBW	Normal Birthweight
EPT	Extremely Preterm
MPT	Moderately Preterm
VPT	Very Preterm
FT	Full Term
PT	Preterm
RDS	Respiratory Distress Syndrome
MM	Motor Milestones
ASQ-II	Ages and Stages Questionnaire, second edition
BSID	Bayley Scales of Infant Development
PDI	Psychomotor Developmental Index
PPD	Postpartum depression
IMQ	Infant Motor Quotient (now ASQ)
IMP	Infant Motor Profile
PDMS	Peabody Developmental Motor Scales
DAIS	Daily Activities of Infants Scale
AHEMD-IS	Affordances of the home environment - Infant-Scale
DDST	Denver Developmental Screening Test
TBCS	Taiwanese Birth Cohort Study developmental instrument
MSEL	Mullen Scale of Early Learning
M-ABC	Movement-ABC
MTM	Motivation to Move scale

NBAS	Neonatal Behavioral Assessment Scale
NPI	Neonatal Perception Inventory
EPDS	Edinburgh Postnatal Depression Scale
SF-36	36-item Short Form Health Survey
KIDI	Knowledge Infant Development Inventory
CP	Cerebral Palsy
PBs	Parental Beliefs
SES	Socioeconomic status
N/A	Not Applicable
LGM	Latent Growth Model
LMM	Linear Mixed Model
ICC	Intraclass Correlation Coefficient
SEM	Standard Error of the Measurement
SDC	Smallest Detectable Change
M	Mean
SD	Standard Deviation

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ABOUT THE AUTHOR

Curriculum Vitae

Marike Boonzaaijer was born in Glimmen (Haren, Gr.), the Netherlands, on September 9th 1975. In 1992, she received her VWO-diploma at the Johannes Fontanus College in Barneveld. She completed her Physical Therapy study at the HU University of Applied Science in Utrecht in 1996 and started working at the St. Antonius Hospital Utrecht (Overvecht and Ouderijn). One year later, she started her Master in Educational studies, specializing in children with motor disabilities at Utrecht University. In 2000, she received her Master of Science degree with a thesis on motor development and quality of life after epilepsy surgery at the Wilhelmina children's hospital (WKZ/ UMCU), Utrecht. After this, she combined her work at the St. Antonius hospital with a specialization in Pediatric Physical Therapy. In 2006, she received her diploma at the HU University of Applied Science, Utrecht. Besides working in the Pediatric and Neonatal Departments at the St. Antonius hospital (Nieuwegein), Marike also started to teach at the Master Physiotherapy, specialization Pediatrics (HU) in 2013. The same year, she participated in research within the GODIVA project (Gross mOtor Development of Infants using home Videos with the AIMS). In 2016, she received a personal teachers grant from the Netherlands Scientific Organization (NWO) to start a Ph.D. about the variability in gross motor trajectories of term-born infants. While finalizing her Ph.D. in 2020, Marike accepted the opportunity to work at the Neonatal follow-up clinic at the WKZ/UMCU, Utrecht. In the future, Marike aims to continue her research on early motor development at the HU University of Applied Science, Research group Lifestyle and Health, to connect this research to both PPT education and PPT practice. Marike Boonzaaijer is married to Joost Brouwer. Together they have three children: Bram (2003), Floor (2006), and Viktor (2009).

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