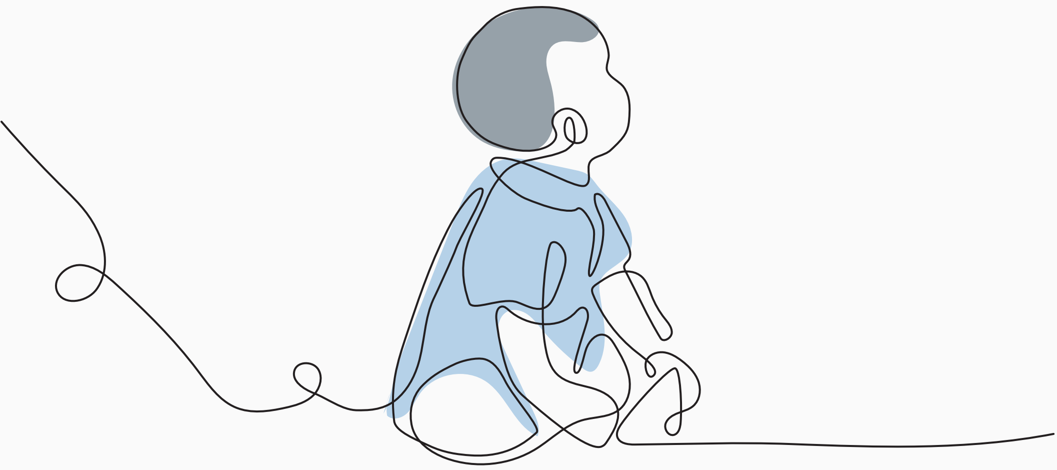
Step by Step

Gross motor trajectories of very preterm infants until reaching the milestone of independent walking



Imke Suir

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Stap voor Stap

Grofmotorische ontwikkelingstrajecten van veel te vroeg geboren kinderen tot zij los gaan lopen

(met een samenvatting in het Nederlands)

Proefschrift

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A journey of a thousand miles, begins with a single step.

(Lao-Tse, Chinees filosoof +/- 600 BC)

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Chapter 1

General Introduction

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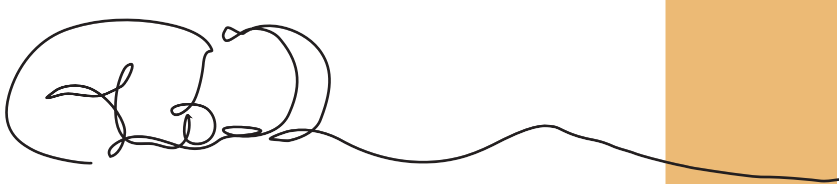

General Introduction

Parents are seeing a paediatric physiotherapist (PPT) for the first time, with their 5-month-old premature infant. They are a bit nervous, because their infant was born at a gestational age (GA) of 29 weeks, a birthweight (BW) of 1356 grams, and a five-minute Apgar score of 6. It was a hectic start, with concerns about their infant's health. Their son was admitted to the Neonatal Intensive Care Unit (NICU) for 10 weeks and parents were involved in the daily care. Once at home, it all became easier, and their infant seems to be a happy and quiet baby. The neonatologist told them that their infant would benefit from some support in its motor development, because during the most recent visit to the follow-up clinic he did not yet lift its head when laying on his belly. Parents told the neonatologist they never put him on its belly. He is the parents' first child, and they do not know what to expect from the progress of their infant's motor development. They have a neighbour from Afghanistan with a baby, who has approximately the same (calendar) age, and is already turning from her back to her belly, However, they know they should not compare their infant with their neighbour's baby, because, when corrected for premature birth, their baby is only 9 weeks old. The neonatologist recommended visiting the PPT / ToP [1] PPT, who has an education specialized in recognizing health problems that affect movement from infancy through adulthood. They hope the PPT will confirm that their infant is doing well and eventually will get to independent walking.

[1]ToP: After discharge from the hospital, infants born before 32 weeks GA or with a BW less than 1500 grams are advised to participate in the ToP program (Transmural developmental support for preterm born infants and their parents), a post-discharge responsive parenting program for preterm born infants and their parents performed at home by trained pediatric physiotherapists, covered by the Dutch health insurance system [7].
Throughout the thesis in the presented cases, PPT will be used to refer to either a ToP PPT or a regular or general PPT.

This case illustrates the daily clinical practice of a paediatric physiotherapist. In fact, it addresses topics that this thesis focuses on, like preterm birth, variation in gross motor development, factors that influence (premature) infant gross motor development, and lastly parental beliefs and practices. By gaining insight into these topics, this thesis aims to contribute to clinical decision-making of paediatric physiotherapists together with parents, and with that shape early intervention.

Premature birth

Premature birth and nationwide monitoring of development

Already in the 17th and 18th centuries, literature exists about neonatal interventions, given that the mortality rate of premature infants was extremely high. During the last decades of the 20th century, substantial medical progress has been made, resulting in the survival of infants being born after 23 weeks of gestation [1]. With the increase in survival rate, challenges remain in improving the neurodevelopmental outcomes of premature infants [2].

In the Netherlands, 6.7% of the approximately 164 000 infants born in 2020, were preterm, of whom 0.5% extremely premature (<28 weeks GA) and 0.6% very premature (28-32 weeks GA) [3,4]. When an infant is born extremely or very premature, the infant is admitted to the NICU. The less mature the infant, the more the infant is at risk of a variety of complications, such as brain lesions (intraventricular haemorrhage and white matter injury), abdominal problems (necrotizing enterocolitis), infections (sepsis, meningitis), and lung problems (bronchopulmonary dysplasia). During this period, it is important to involve parents in the medical decisions made and daily care of their infant, by providing family-centred care [5].

After an oftenlong period of hospitalization, the infant is discharged, and parents can take their infant home. In the Netherlands, from that time onward, the infant is monitored according to the protocol agreed upon by the nationwide Dutch Neonatal Follow-Up (LNF) Study Group for infants admitted to the NICU, during regular visits to a follow-up clinic [6]. Infants visit the clinics spread across the country at the ages of 6, 12, 24 months corrected age (CA; i.e., age calculated from the actual scheduled time of birth), and 5 and 8 years [6,7]. During the regular check-ups, standardized tests are conducted which cover the surveillance of several developmental domains like neuromaturation, motor development, cognitive development, behavioural development, and executive functions. For the motor development at the check-up at 6 and 12 months CA, the Alberta Infant Motor Scale (AIMS) is usually administered.

It is important that premature infants are monitored, because they are at risk of neurodevelopmental problems, like problems related to gross and fine motor development, cognition, and/or behaviour [8,9]. Approximately 20% of infants experience an overall motor delay at around 2 years CA, with a prevalence of 36% at preschool age and persisting into adolescence [2,10]. Early detection of developmental problems enables early interventions to support neurodevelopmental outcomes during infancy, and into preschool age [2,9,11–13].

Measuring infant motor development

Motor development is an early marker in the first year of life to identify developmental delay [9,14]. Difficulty in identifying delay lies in the variable course of motor development in infants, which makes predicting infant motor development, and with that, clinical decision-making difficult [15–18]. Measuring motor development makes discrimination of typical or atypical development and developmental progress insightful. Clinicians need these measuring instruments for clinical decision-making and shaping early interventions. Moreover, due to the intra- and interindividual differences in developmental speed and course of development, it is necessary to measure an infant more than once over a period of time [16,19]. There are several measurement instruments available for objectifying gross motor development, like the Bayley Scales of Infant Development (BSID-II-NL) [20], the Peabody Developmental Motor Scales 2nd Edition (PDMS-2) [21], the Infant Motor Profile (IMP) [22] or for distinguishing normal from abnormal general movements the General Movement Assessment (GMA) [9,23]. Also registering the age of attaining a motor milestone -rolling from supine to prone, independent sitting, crawling and independent walking- is an easy, but rather crude way of assessing gross motor development.

Alberta Infant Motor Scale (AIMS)

A world-wide used measure for gross motor development is the AIMS [24–35]. It is (also) the preferred measure to be administered at the ages of 6- and 12-months CA when infants visit the Dutch neonatal follow-up clinics (see above). The AIMS is used as a main outcome measure in this thesis.

The AIMS is an observational instrument to examine, discriminate and evaluate the spontaneous gross motor repertoire of the infant from birth until independent walking in four different postures: prone, supine, sitting, and standing position. It is a valuable tool for infants at risk of neurodevelopmental problems, as it is able to detect deviations in motor maturation [14,36,37]. Motor maturation is characterized not only by motor milestones but also by qualitative aspects of motor performance, which are captured in an AIMS assessment [36]. Each of the 58 items of the AIMS is a small motor milestone that is characterized by qualitative aspects which can be observed by the assessor. A window of the current skills is represented by the least and most mature observed items in each position and represents the motor repertoire of the infant [37].

For a trained professional the assessment is relatively easy. Administering only takes a short time. The AIMS has good psychometric properties with high intra- and interrater reliability and has norm references which were established in 1994 in Alberta (Canada) [14]. In 2014, a re-evaluation of the norm references was conducted given changes in postnatal policy (e.g., back to sleep campaign) and a growing ethnic diversity of the Canadian population since 1994. The results of the re-evaluation revealed that after 20 years, the norm references remained valid [38]. Despite this, there were still questions about the cross-cultural validity of standardized gross motor developmental screening and assessment tools. Several studies show that there are ambiguous results regarding the validity of the AIMS norm references in other countries. For example, in Greece there seem to be no differences in the AIMS values compared to the Canadian norm references [25]. However, in Brazil and Belgium, there seems to be a difference in developmental pace, where both Brazilian and Belgium infants develop slower than Canadian infants [29,30]. In a Dutch pilot study by Fleuren et al. a similar trend was reported. Dutch infants seem to develop more slowly compared to Canadian infants assessed with the AIMS [31].

Also, for infants at risk, the AIMS is a valid and reliable tool for assessing gross motor development [39,40]. When administering the AIMS, premature infants are corrected in age (CA) for their prematurity. Research has already confirmed that premature infants show inferior gross motor performance in the first 18 months (CA) of life [36,41].

AIMS home-video method

In 2013, the GODIVA research project (Gross mOtor Development of Infants using home Video registration with the AIMS) started within the Research Group Lifestyle and Health of the Utrecht University of Applied Science together with many other partners within a consortium, like PPTs from primary practices and hospitals, the Faculty of Computer Engineering (Utrecht University of Applied Science), and the Faculty of Social and Behavioural Sciences (Utrecht University). The aim of the project was to explore the possibility of remote monitoring of young infants' motor development. The result was the development of a new application for the AIMS, the AIMS home-video method [42].

With this new method, parents record their infant according to specific instructions, which consists of three instruction videos and a booklet with three corresponding checklists **(see Appendix I)** on how and what to record and how to upload recordings on a secure digital server. Parents recorded their infant in the four AIMS positions at a convenient time and place for both parents and infant. After uploading the recordings, the AIMS was assessed by the researchers/PPTs. Parents were given feedback about their infants' motor development via e-mail [43].

The home-video method appeared a valid and reliable measurement in the assessment of the AIMS, with an intra-class correlation agreement between live and video assessment of .99 and a standard error of 1.41 items [42]. Also, the AIMS home-video method was considered feasible for parents of typically developing, full-term (FT) infants to record their infant five times with a two-month interval between each video [44]. This new home-video method, therefore, was regarded as a valuable approach also for studies reported in this thesis in monitoring premature infants longitudinally for research purposes and potentially also for clinical use. Besides, it provides the opportunity to follow infants and gain more insight into the individual variability of gross motor trajectories.

Factors influencing infant gross motor development

Infant factors

The intra- and inter-individual variability in (premature) infant gross motor development may be explained by the influence of many factors. According to the Dynamic Systems Theory (DST) framework for infant motor development of whom Esther Thelen was its founder, the DST principles are based on the belief that motor skills emerge as a result of the self-organization of an array of parameters within the child, the task and the environment [17]. Besides, according to Esther Thelen, each child does not mature at the same rate or in the same linear pattern. Due to the interaction of the child, the task and the environment, changes in one of these subsystems can result in new motor behaviour [17,45].

This variability in motor development is seen in the great variation regarding the age of attainment of gross motor milestones, like independent sitting, crawling, and walking. Many factors contribute to this variability in attainment [45]. A tremendous amount of research has been conducted on the factors influencing infant development in general. Most of this research is on infant characteristics, like GA, BW, sleeping position, and the use of equipment (e.g., a baby walker) [2,10,36,46,47]. However, this research mainly comprises cross-sectional research designs. Due to the variability in development of infants, longitudinal studies are preferred following children over time, providing more information about factors associated with infant motor development [48].

Environmental factors: parental beliefs and practices

Next to infant characteristics, there are also environmental factors contributing to gross motor development, like the direct surroundings of the infant. During the first years of life an infant depends directly on its parents or caregivers who facilitate infant development by providing a rich environment in which infants can explore and learn.

Those environmental factors, mostly facilitated by its parents or caregivers, may promote or hinder motor development. An example is the use of an occluding baby walker (a baby walker in which the infant is not able to see its own feet) which hinders motor development, resulting in delayed gross motor development between 6 and 15 months of age [47,49,50]. Sleeping in prone position on the other hand is promoting gross motor development [47].

According to the Developmental Niche, parental practices are influenced by parental beliefs. The Developmental Niche, with Super and Harkness as its founders, is a theoretical framework of infant development based on ethnocultural research [51,52]. The framework depicts an interaction between the infant/child, it's physical and social setting, caregiver psychology - e.g., parental beliefs about development and parenting-, and the socio-cultural context of the child and its parents. The social-cultural context represents the daily customs and childrearing practices [52,53]. Beliefs serve as a basis to understand the world around us and therefore are held to be true. They have different origins and can be conscious as well as unconscious. Information from a trusted source, but also past experiences may form beliefs [54]. Parental practices influence infant motor development [55,56]. The hectic start of preterm birth can determine the beliefs parents have about what to expect of their infant [57,58]. For example, mothers offer more immature toys to infants who are premature-labelled [58]. This reflects the interaction between parental beliefs and parental practices. Yet, not much is known about the beliefs of parents of Dutch premature infants, while culture also plays an important role in the parental beliefs and practices [51,53,59].

One of the ways in which to measure parental beliefs regarding motor development is by administering the Parental Beliefs on Motor Development (PB-MD) questionnaire [60], a measure chosen to be included in this thesis. The questionnaire is based on the framework of the Developmental Niche.
It consists of five scales measuring the beliefs that:

1. stimulation of motor development is important
2. motor development occurs naturally
3. seeking advice on motor development is important
4. order of motor development is important
5. children should follow their own pace in motor development

Besides, questions are asked about how important parents think motor development is in the first year of life, and two open questions about the role parents have in motor development and what activities they provide for their infant's motor development. The psychometric properties of the questionnaire are good [60].

Aims and outline

This thesis is a continuation of the GODIVA research project. To make its unique focus clear the study is called the GODIVA-PIT (following Premature Infants in Time) study. It comprises three main parts with each their own focus:

Part I: Infant and environmental factors

Investigating factors influencing infant gross motor development until independent walking.

Chapter 2: With the AIMS-home video method, it was possible to obtain a large amount of data. Because next to cross-cultural research into the AIMS accompanied by the voiced concern of Dutch PPTs that Dutch infants are more often identified with a developmental delay according to the Canadian AIMS norm references than they would identify based on clinical views and expertise, the AIMS-NL study started. This study had the aim to assess the cross-cultural validity of the Canadian (2014) norm values of the AIMS evaluated for Dutch infants.

Chapter 3: Due to the great variability in the attainment of gross motor developmental milestones, there is a need to understand which infant and environmental factors influence motor development. Knowledge of these factors may help clinical decision-making and shape interventions. Therefore, a systematic literature review was conducted to provide an overview of infant and environmental factors associated with gross motor development from birth to independent walking, solely based on longitudinal studies.

Part II: Infants

Exploring gross motor developmental curves of very premature (VPT) infants or infants with BW < 1500 grams from 3 to 18 months CA without severe perinatal complications.

Chapter 4: Although the AIMS is a valid and reliable measure for gross-motor development, accuracy is limited with only one assessment, because every infant develops in its own way and speed. Therefore, it is important to gain insight into individual motor trajectories and thus it is necessary to study longitudinal data. With the AIMS-home video method, it became easier to collect data independent of time and place and it became possible to follow infants for a longer period. This study firstly aimed to explore gross motor developmental curves from 3 until 18 months CA and its related factors among VPT infants. Furthermore, it aimed to explore whether separate motor profiles can be distinguished and compare these to profiles of Dutch full-term infants.

Part III: Parents

Examining parental experiences with the AIMS home-video method and parental beliefs and role regarding gross motor development of VPT infants.

Chapter 5: Nowadays, eHealth technology is becoming more common in daily practice. Moreover, the need for remote healthcare became extremely relevant during the recent COVID-19 global pandemic. The AIMS home-video, although developed for research purposes, may be an additional method to facilitate care and guidance, with VPT infants returning to the hospital for regular check-ups. A qualitative study was conducted to gain an understanding of the experiences of parents of VPT infants in using the AIMS home-video method and how they would appraise its applicability for use in an outpatient follow-up clinic.

Chapter 6: Infant motor development emerges from the interaction between the infant and its environment (e.g., its parents). Parental practices are influenced by the beliefs parents have about infant development. Little is known about parental beliefs regarding motor development. Therefore, this study explored the similarities and/or differences in parental beliefs regarding motor development between Dutch parents of VPT and full-term (FT) infants, measured with the PB-MD questionnaire. Besides this, differences in how parents of FT and VPT infants consider their own supporting role in the motor development of their infant are examined.

The purpose of exploring and examining gross motor development of Dutch (premature born) infants, is to offer evidence-based knowledge contributing to early detection of gross motor delay, clinical decision-making and interventions. More knowledge about individual gross motor developmental trajectories and factors that influence these trajectories will feed more realistic expectations of parents and PPTs. Also, knowledge about parental beliefs regarding gross motor development and the role parents think they have, may help PPTs in understanding parental considerations and may help align interventions to parental needs and beliefs. All to improve health care for parents and (premature) infants.

With these insights, the PPT who is being visited by parents in the earlier presented case will have tools to objectify whether this Dutch infant is lagging behind, compared to Dutch full-term and/or preterm peers. Besides, by knowing the parental beliefs of these parents, the PPT is able to understand why parents do not put their infant on its belly, which is the starting point of the intervention.

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Appendix I: Checklist I-II-III

Checklist I The baby is not rolling over yet

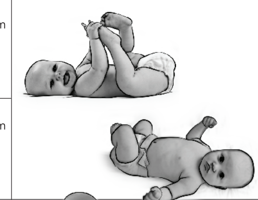
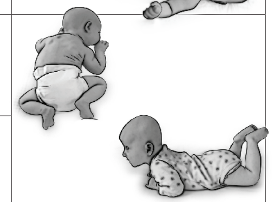
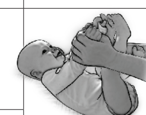
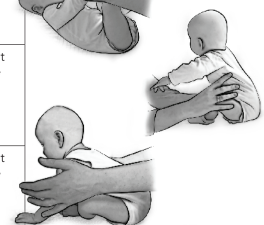
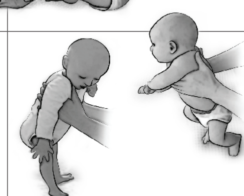
This checklist can be used during filming. Don't forget to watch the instruction video.
In this checklist you will find:
- The movements and positions we want you to capture on the home video.
- Tips to pay attention to, so your home video can be used to assess motor performance.

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.

[1]The development of this checklist was part of a grant reseach project.
[2]To enhance the ease of use for parents, some changes in the lay-out were made.

Checklist I The baby is not rolling over yet

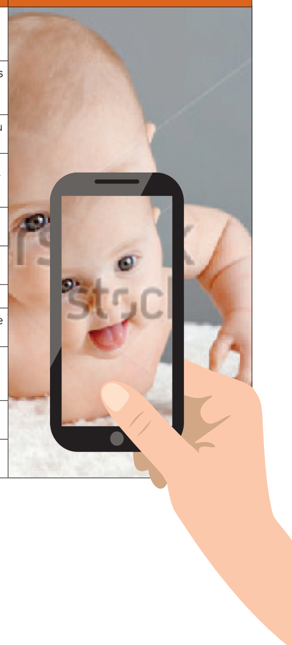
Position	Check the tips	Camera position from the
Supine	☐ Film a few minutes in supine position without a toy.	side bottom
	☐ Present a little toy above your baby, to elicit reaching and/or grabbing.	side bottom
Prone	☐ Lay your baby down in prone position with his/her hands at shoulder level. Film a 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.

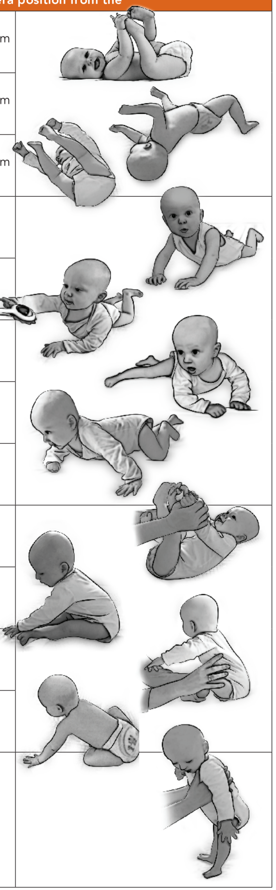
Check	Tips
General	☐ We will assess the motor skills of your baby, so let him/her move freely and try not to help with your hands.
	☐ A good way to start the video is to film spontaneous movements of your baby; please don't elicit movements with toys or sounds right away.
	☐ During filming, make contact 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.



[1]The development of this checklist was part of a grant reseach project.
[2]To enhance the ease of use for parents, some changes in the lay-out were made.

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	side bottom
	☐ Present a toy beside the head of your baby, maybe he/she will roll over	side bottom
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.	side top
	☐ 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.	top side
	☐ 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.	top side
	☐ 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.	front side
	☐ 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, it is better to stop and try filming again another time.

[1]The development of this checklist was part of a grant reseach project.
[2]To enhance the ease of use for parents, some changes in the lay-out were made.

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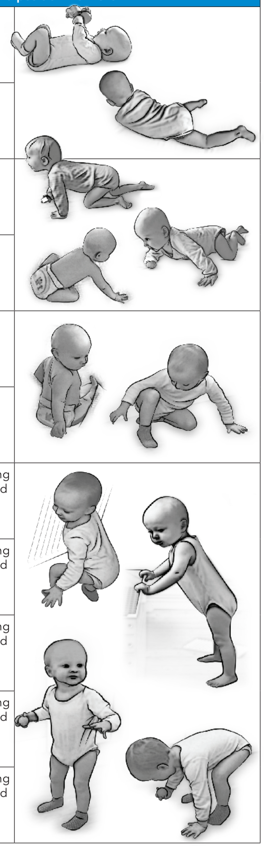
Checklist III The baby is crawling and / or walking

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.	side
Prone	☐ Capture your baby moving forward, this can be crawling or creeping.	side
	☐ Try to film the transfer from sit to crawling.	side
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.	side
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.	walking around
	☐ Is your baby capable to transfer from a standing position to a sitting position? Try to capture this.	walking around
	☐ If your baby is capable to stand or walk without support, try to capture this.	walking around
	☐ Playing in a squatted position without support is the final position to film.	walking around



PART I

Infant and environmental factors

Chapter 2

Chapter 3

Part I

Infant and environmental factors

Chapter 2

Cross-cultural validity: Canadian norm values of the Alberta Infant Motor Scale evaluated for Dutch infants

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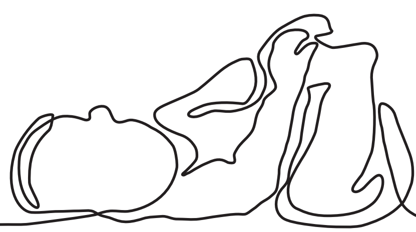


Abstract

Purpose: To examine whether the currently used original Canadian normative values of the Alberta Infant Motor Scale (AIMS) are appropriate for infants in The Netherlands.

Method: The AIMS-NL study was a replication of the 2014 re-evaluation study of the Canadian normative data including typically developing infants between two weeks and 19 months. Parents used the AIMS home-video method to film their child.

Results: 499 Dutch infants were included (7.6% premature and 5.8% infants of non-Western origin). Of the 45 AIMS items which met the criterion for stable regression to calculate item locations, Dutch infants passed 42 items at a later age than the Canadian infants. Almost all monthly age groups of Dutch infants showed significantly lower mean AIMS scores.

Conclusions: The Canadian norms seemed not to be appropriate for the Dutch study sample. These findings have several implications for clinical use and research regarding the AIMS in The Netherlands.

Key words: AIMS, Dutch, infants, norm values

Introduction

The Alberta Infant Motor Scale (AIMS) is a well-known and frequently used observational instrument to measure infant gross-motor development from birth until independent walking (0-19 months). The AIMS allows clinicians to discriminate typical from atypical motor development and was originally developed and norm referenced with 2202 infants in Alberta, Canada in the early 1990s [1]. The neuromaturational theory provides a framework for the infant's sequential motor development assessed with the AIMS. Variability in rate of acquiring motor skills is better explained by the dynamic systems theory, where infant development is considered in its context [2]. Because of these contextual influences, the question arose whether policy (e.g., back to sleep campaign) and changes in ethnic diversity of the Canadian population would have influenced infant motor development.[3] The re-evaluation study published in 2014 provided evidence that in 20 years the established norms for the Canadian infants remained valid [3].

In their review, Mendonca et al. (2016) investigated the cross-cultural validity of standardized motor developmental screening and assessment tools to evaluate motor development of children up to two years of age [4]. They concluded that standardized motor developmental assessments have limited validity in cultures other than the culture in which the normative sample was established. The use of culturally specific assessment tools might have consequences for clinical use, in terms of under- or over-referral for services.

Several studies examined the validity of the AIMS normative values in other countries [5-7]. Ambiguous results were reported. Syrengelas et al. found no difference between Canadian and Greek infants in a sample of 1068 full term born Greek infants [5]. On the other hand, a cross-cultural analysis of motor development of Brazilian, Greek and Canadian infants assessed with the AIMS showed that Brazilian infants are developing more slowly according to the AIMS than the Greek and Canadian infants [6]. A Dutch pilot study by Fleuren et al. reported a similar trend. Dutch infants seem to develop more slowly compared to Canadian infants assessed with the AIMS [8].

The results of these reports are accompanied by the voiced concern of pediatric physical therapists (PPTs) in The Netherlands. They notice that Dutch infants are more often identified with a developmental delay according to the Canadian AIMS norms, than they would identify based on their clinical view and expertise.

The contextual influence of culture on normative values still remains a discussion [9]. Moreover, this makes a study to identify whether the Canadian norms are valid for the Dutch population relevant. Therefore, the objective of this study is to examine whether the currently used original Canadian AIMS norms are appropriate for infants in The Netherlands.

Method

The AIMS-NL study was a cross-sectional descriptive study, comparable to the re-evaluation study of the Canadian norm values of 2014 [3]. The Research Ethics Committee of University Medical Center Utrecht approved the study (protocol nr.15-029C).

Participants

As in the Canadian re-evaluation of the AIMS norms, the target was to include a minimum of 450 Dutch infants, with an overall spread over the monthly age groups of two weeks to 19 months **(see Appendix I)**. This spread was in proportion with the original AIMS distribution. Infants with pathology or a disorder known to influence gross motor development were excluded. Ideally, the sample should comprise approximately 8% premature born infants (gestational age (GA) <37 weeks) and 10% infants from non-Western origin, which is in accordance with the population composition in The Netherlands [10]. Confirming to Statistics Netherlands (www.cbs.nl), non-Western origin is defined as an infant either having at least one parent (first-generation) or grandparent (second-generation) from non-Western origin [11].

Under the umbrella of a long-term research project named GODIVA, data was collected in multiple studies. GODIVA is an acronym for Gross mOtor Development of Infants using home-Video registration with the Alberta Infant Motor Scale. The GODIVA project started in 2013 and is still running. The GODIVA project comprises five studies, including the current AIMS-NL study. Two studies, namely the AIMS home-video validation study [12] and a pilot longitudinal study on gross motor development in infants between 1.5 and 15.5 months are completed. The pilot longitudinal study explored the feasibility of the home video method for parents in longitudinal research design.[13] To date, two other longitudinal studies are still running. All these studies generated cross-sectional data for the AIMS-NL study. In addition, participants were recruited specifically for the AIMS-NL study between January 2014 and April 2018. For all GODIVA studies recruitment of infants was conducted via social media, word of mouth, flyers, and posters in Well Baby Clinics. After showing interest to participate, parents received official information and video-instructions together with the Informed Consent form they all signed.

Measurements

The AIMS is a norm-referenced observational instrument which measures infant gross motor development (0-19 months) and has good psychometric properties [1]. The infant is observed in four different postures (supine, prone, sitting and standing). The test items are mainly based on spontaneous movements of the infant. The instrument consists of 58 items, which can be scored as: 'observed', 'not observed' or 'mastered'. The observed items together represent the actual motor repertoire of the infant [1].

Test procedure

To collect data, parents used the AIMS home-video method. This method is a validated and reliable way of assessing the AIMS with an ICC agreement between live- and video-assessment of 0.99 and standard error of measurement (SEM) of 1.44 items.[12] In this method parents are asked to film their child in the four positions of the AIMS with guidance of an instruction film and instruction card. Using this method ensures that the infant is in its own environment and filmed at a convenient time for both parents and child. After recording, parents uploaded the home-videos in a secure digital environment or saved it on a secured USB-stick. The researchers scored the AIMS from the video recordings and parents received feedback by e-mail.

Most videos were assessed by two trained PPTs/researchers. The agreement on item level between the two observers on eight infants was 97.8%. Consensus meetings were organized frequently to discuss disputable items. In total, four other trained PPTs assessed AIMS home-videos. Their training consisted of two four-hour-training sessions where infants were scored from videos. At the end of the training, two videos were scored and they passed the training when an item agreement of 80% with the consensus score of three experienced researchers was obtained.

Data analysis

Participant characteristics were calculated using descriptive statistics. Depending on type of measurement, the mean and standard deviation (SD), median with interquartile range (IQR) or counts with percentage (%) are presented. The significance level was set to 5% and all data was analyzed using the statistical package SPSS 24.0.

Data from the original Canadian normative dataset was compared with Dutch data using the scaling method [3]. In this method an estimate of the age at which 50% of the infants would pass an item, is referred to as item location. For each item of the AIMS, logistic regression was used to calculate the item location. The second step was to compare those items of the Canadian- with the Dutch sample, which had a proportion of infants passing the item between 0.10 and 0.90 in both samples. Of the proportionally valid items, the means and SD of the item locations were calculated in the third step. The last step in the scaling method was to plot for each item the item location of Dutch infants against the Canadian infants and a regression model was fitted. As we assumed that the AIMS-score of an infant starts at zero, therefore a regression model with intercept zero was fitted.

If the order of the items does not change, then a curvilinear transformation of the original scale would be necessary to generate new tables. To objectify this, the mean total AIMS score had to be compared between Canadian and Dutch infants with the Welch's t-test. The mean total AIMS scores were plotted and the centile rankings (P5, P50 and P90) were presented.

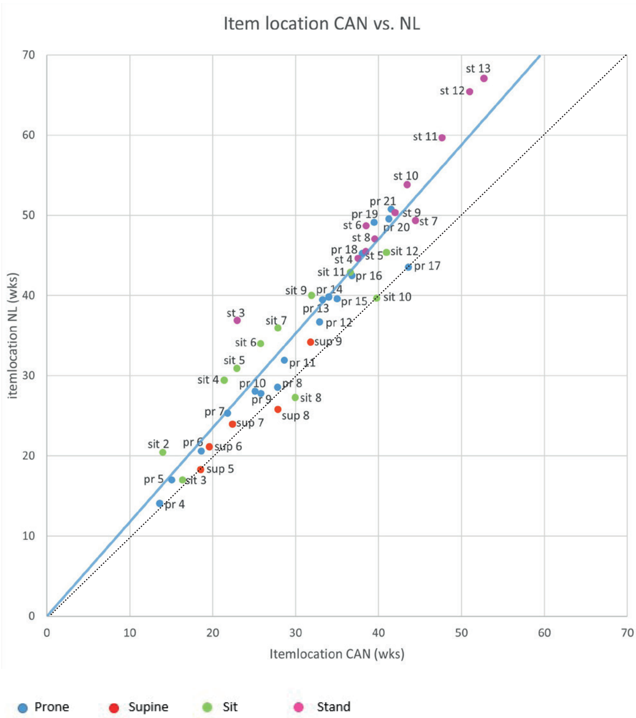
Results

A total of 499 infants participated in the study of which 263 were boys (53%). Furthermore, 38 infants (7.6%) were born preterm with a mean GA of 31.2 weeks (SD 3.5), 27 with GA of <34 weeks and 11 between 34-37 weeks. Infants' mean test-age was 38.2 weeks (SD 18.9) in comparison with 37.4 weeks (SD 17.6) in the original Canadian sample (p=0.39). Of 412 participants 5.8% were of non-Western origin. The age of 170 mothers and 153 fathers was known, with a modal age of 30-35 years (43% mothers and 39% fathers). Almost 80% of the 234 mothers and 229 fathers were highly educated.

Results of the scaling method demonstrating the item location are presented in **Appendix I**. Thirteen items of the 58 items were removed from analysis, due to homogeneous scoring or because the proportion of infants passing the item was below 0.10 or above 0.90 in one or both data sets. The remaining 45 items were included in the analysis. In 42 items Canadian infants show a younger mean age of passing an item. The biggest difference was 14.42 weeks for early stepping (item Stand 12): Canadian infants passed this item at a mean age of 50.99 weeks (11.8 months), and Dutch infants 65.41 weeks (15.1 months).

Items Supine 5 (hands to knees) and Supine 8 (rolling to supine without rotation) are passed at a somewhat younger mean age by Dutch infants, with a two-days and two-weeks difference respectively. Items Sit 10 (sitting to prone) and Supine 17 (reciprocal creeping 1) are observed at nearly the same mean age, with -0.07 and 0.01 difference.

When item locations of the 45 items are plotted **(Fig. 1)**, a linear line $X_{Dutch} = -7.42 + 1.21 * X_{Canada}$ is fitted with a proportion explained variance (R^2) of 92.7%. If it is postulated that the intercept is zero, then the linear regression model is $X_{Dutch} = 1.18 * X_{Canada}$ with a 7% higher proportion explained variance (R^2= 99.2%).



reference line where CAN = NL

linear line with intercept = 0

Figure 1: *Difference of the item location for the Dutch infants compared to the Canadian infants on the mean age (weeks) scored per item.*

Distribution of the Dutch monthly age groups in comparison with the Canadian original normative data is presented in **Table 1.** Most differences between the Dutch and Canadian infants were significant, except for infants of 3-4 months old and 17-19 months.

Table 1: Distribution of monthly age groups of the original Canadian and the Dutch sample

Age group (months)	n NL/CA	Boys/girls	Mean (SD) total Dutch AIMS scores	Mean (SD) total Canadia AIMS scores n	Difference Difference CAN-NL	*p* value (Welch's test)
0-1	10 / 22	6 / 4	5.6 (1.17)	4.5 (1.37)	-1.1	0.03*
1-2	13 / 56	9 / 4	6.7 (1.49)	7.3 (1.96)	0.6	0.23
2-3	21 / 118	9 / 1 2	8.6 (1.69)	9.8 (2.42)	1.2	0.01*
3-4	30 / 90	14 / 16	12.3 (2.55)	12.6 (3.29)	0.3	0.61
4-5	27 / 122	21 / 6	14.9 2.66)	17.9 (4.15)	3.0	0.00*
5-6	44 / 189	20 / 24	18.5 (4.22)	23.2 (4.75)	4.7	0.00*
6-7	41 / 225	22 / 19	22.4 (3.40)	28.3 (5.50)	5.9	0.00*
7-8	44 / 222	22 / 22	28.8 (7.43)	32.3 (6.85)	3.5	0.01*
8-9	42 / 220	24 / 18	31.9 (7.85)	39.8 (8.69)	7.9	0.00*
9-10	44 / 189	22 / 22	37.0 (8.83)	45.5 (7.47)	8.5	0.00*
10-11	33 / 155	21 / 12	43.0 (7.72)	49.3 (5.92)	6.3	0.00*
11-12	31 / 155	15 / 16	44.5 (8.37)	51.3 (7.11)	6.8	0.00*
12-13	29 / 124	15 / 14	50.2 (6.55)	54.6 (4.52)	4.4	0.00*
13-14	20 / 86	13 / 7	51.5 (4.86)	55.6 (5.01)	4.1	0.00*
14-15	21 / 61	10 / 11	51.8 (4.98)	56.9 (1.97)	5.1	0.00*
15-16	18 / 40	9 / 9	56.4 (1.94)	57.8 (0.45)	1.4	0.01*
16-17	13 / 49	5 / 8	54.5 (4.50)	57.8 (0.55)	3.3	0.02*
17-18	9 / 49	4 / 5	57.3 (2.00)	57.9 (0.35)	0.6	0.40
18-19	9 / 30	2 / 7	56.7 (2.50)	57.7 (0.64)	1.0	0.27
Total	499 / 2202	263 / 236				

**significant when α <0.05*

Figure 2 demonstrates the centile distribution of AIMS scores of the Dutch and Canadian infants. A similar course of the distribution is seen, with Dutch infants having lower scores in all age groups except the first (<1 months). At 16 months a decline in Dutch AIMS score is detected, which might be explained by one premature infant with a total-score of 42 points. Compared to the Canadian centile curves, 74% of the Dutch infants scored below the 50th centile of which 16% of the infants scored below the 5th centile. According to the Canadian normative values, these infants would have been classified as having a motor delay.

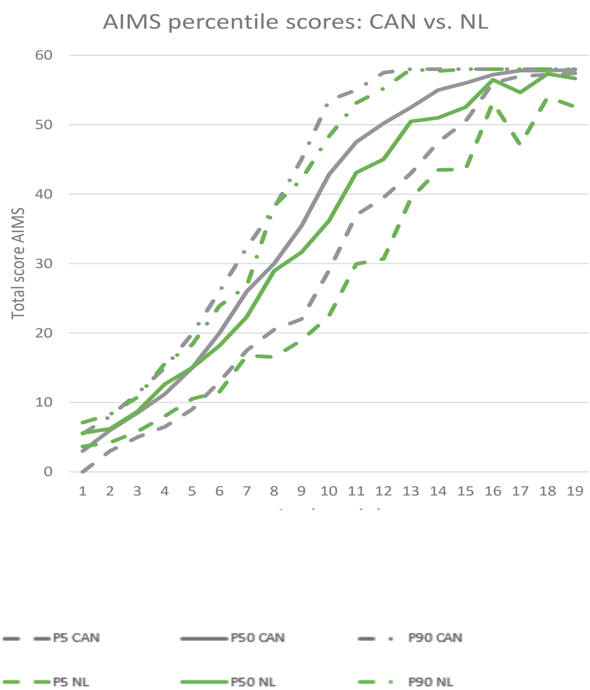


Figure 2: AIMS percentile scores (P5, P50 and P90) of the Canadian and Dutch infants.

Discussion

The AIMS-NL study is the first study in which the item locations of the AIMS items are calculated and compared with the Canadian item locations with the purpose to examine whether the currently used original Canadian AIMS norms are appropriate for Dutch infants. The study revealed that the mean age at which 50% of the infants pass an AIMS item is at a later age than the Canadian norm group. Regardless of differences in rate between the Dutch and Canadian infants, the AIMS-NL study also demonstrates that the sequence of the AIMS items did not change, which means that infants attain motor milestones in the same developmental order as the Canadian infants. This is in support of the theoretical construct of the AIMS, the neuromaturational theory, where the sequence of the individual motor items is considered equal for the majority of infants.[2]

Fifty percent of the Canadian infants passed almost all items at an earlier age. Only three items are reached earlier by Dutch infants. Surprisingly item Prone 17 (reciprocal creeping) is passed at the exact average age by the Dutch and Canadian infants. Remarkably, in the original AIMS norms reciprocal creeping with rotation (item Prone 21) is passed at an earlier age (41.49 wks) than reciprocal creeping without rotation (Prone 17; 43.56 wks), while in the Dutch sample, infants pass item Prone 21 at 51.19 wks and Prone 17 at 43.57 wks, which seems a more logical order. Item Prone 21 was removed in the Canadian re-evaluation study, because of a large difference in mean age of passing this item. Darrah et al. had concerns this item was scored incorrectly. However, it can be discussed whether the incorrect scoring of the item occurred in the original study or in the re-evaluation study [3].

The findings of our study extend those of a recent study on the Bayley Scales of Infant and Toddler Development (Bayley-III), another measurement tool for infant development [14]. Steenis et al. identified significant differences in the age at which Dutch infants reached gross motor milestones compared to American infants aged 3.5 to 25.5 months. The study confirmed that the item sequence was adequate for the assessment of Dutch children, but more Dutch infants would have been identified with a developmental delay using the American norms of the Bayley-III. Therefore, new Dutch norms were developed [15].

Because the AIMS is never interpreted on an individual item level, it is important to look at the AIMS total scores at various ages. For almost all monthly age groups total AIMS scores differ significantly, except for three age groups around the borders of the test-age, which can be explained by little variances in scores due to a bottom and ceiling effect of the AIMS. According to Darrah et al., the AIMS is considered most sensitive in the period from 4-12 months [2]. As displayed in **Figure 2**, clear differences in total AIMS scores are demonstrated at four months of age. Also, the Dutch 50th centile line is, from four months onwards, below the line of the Canadian norms, whereas 74% of the infants score below the 50th centile. This is in accordance with the results of the pilot studies of Fleuren and De Kegel.[8,16] Fleuren found that 75% of Dutch infants aged 0-12 months score below the 50th percentile of the Canadian norms. De Kegel et al. showed that 64.8% of the Flemish infants score below the 50th percentile.

Strengths of this study are the substantial sample that was included with an adequate representation of premature born infants and the method used, being comparable to the Canadian re-evaluation study to calculate the item locations. The calculated item locations provide a better understanding in the sequence of gross motor development and the differences between the Canadian and Dutch infants. In addition, the results of our study support the PPTs' thoughts about the differences they experience in clinical practice regarding the AIMS scores. Another strength in this study was the usage of the home-video method. The method provided the opportunity for the researchers to discuss difficult items and doing so, increased the reliability of the assessments. This was confirmed by the high agreement (Kappa =0.98) between the two researchers.

There are some limitations to this study that need to be addressed. First, in the age groups 0-3 and 17-19 months, few infants were included. Although this does not affect the item location calculations, it does affect the mean AIMS scores of the age groups. Despite these small groups, the course of the centile scores is comparable with that of the original sample. Additionally, the AIMS has a bottom effect in the earlier ages and a ceiling effect at later ages, therefore the impact of fewer infants in these age groups is less eminent.

Second, it is debatable whether the sample is a perfect representation of the Dutch population. There are missing values for some infant and parent characteristics. In every GODIVA study parents filled out different questions about their demographics. For example, not in every study parental education was asked, resulting in missing data on parental education of almost 50% of the parents. Approximately 80% of more than 200 parents were highly educated. A body of research shows the relation between higher maternal education and better motor outcome [17-19]. Therefore, having a less highly educated sample, would probably not reduce, but rather increase the differences found between the Canadian and Dutch infants. Also, in a smaller part of the infants their origin is unknown. Still, the sample does not count a representable percentage of infants of non-Western origin. The researchers made considerable effort to recruit non-Western parents for the study, with limited results. Language problems, and restraints for filming their child, were some reasons for not participating in the study. Regarding infants of non-Western origin, the results of our study should therefore be interpreted with care.

Despite these limitations, this study reveals relevant differences in developmental rate between the Canadian and Dutch infants according to the AIMS. Future research should therefore explore normative values of other countries then The Netherlands which frequently use the AIMS in clinical practice. In addition, the results of this study raise the question: Which factors contribute to infant gross motor development and the considerable difference in gross motor developmental rate of infants living in different parts of the world?

To conclude, this study reveals that the Canadian normative values of the AIMS do not seem appropriate to evaluate gross motor development in Dutch infants. Dutch infants develop in a similar sequence, but at a slower rate than Canadian infants. This result affects research as well as clinical decision-making based on AIMS data, with the risk of over-referral and perhaps unnecessary parental concern and treatment of infants with gross motor developmental delay. We recommend conducting a study to establish new norm values of the AIMS for Dutch infants based on a larger sample. Until then, we advise PPTs in The Netherlands to take the results of our study into account when reaching a clinical decision based on the AIMS alongside, of course, other available diagnostics as family concerns, medical history and clinical tests.

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AUTHOR CONTRIBUTIONS

Imke Suir: Conceptualization, Methodology, Investigation, Resources, Data Curation, Formal Analysis, Writing – Original Draft, Project administration.
Marike Boonzaaijer: Conceptualization, Methodology, Investigation, Resources, Data Curation, Writing – Review & Editing.
Petra Nijmolen: Conceptualization, Methodology, Investigation, Resources, Writing - Review & Editing.
Paul Westers: Conceptualization, Methodology, Formal Analysis, Writing - Review & Editing.
Jacqueline Nuysink: Conceptualization, Methodology, Writing - Review & Editing, Funding acquisition, Supervision.

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Appendix I: Canadian (original) and Dutch item location of the individual items of the AIMS

Item	Proportion of infants passing the item		Item location Age (wks)			Item not used for equating
	Canada	Dutch	Canada	Dutch	Difference	
PRONE 1#	1.000	1.000				X
PRONE 2	0.979	0.986	3.14	-0.76	-3.90a	X
PRONE 3	0.949	0.950	8.61	7.95	-0.65a	X
PRONE 4	0.907	0.888	13.59	14.08	0.49	
PRONE 5	0.891	0.852	15.05	18.78	1.96	
PRONE 6	0.850	0.804	18.60	20.60	2.00	
PRONE 7	0.796	0.715	21.76	25.38	3.62	
PRONE 8	0.665	0.659	27.79	28.59	0.80	
PRONE 9	0.709	0.671	25.78	27.80	2.02	
PRONE 10	0.736	0.663	25.10	28.06	2.96	
PRONE 11	0.660	0.587	28.62	31.95	3.33	
PRONE 12	0.570	0.505	32.87	36.70	3.83	
PRONE 13	0.562	0.459	33.23	39.51	6.28	
PRONE 14	0.547	0.439	33.99	39.83	5.84	
PRONE 15	0.525	0.445	34.98	39.60	4.62	
PRONE 16	0.486	0.391	36.76	42.50	5.74	
PRONE 17	0.374	0.383	43.56	43.57	0.01	
PRONE 18	0.460	0.353	38.01	45.29	7.28	
PRONE 19	0.432	0.297	39.44	49.13	9.69	
PRONE 20	0.398	0.303	41.21	49.56	8.35	
PRONE 21	0.390	0.273	41.49	50.78	9.29	
SUPINE 1#	1.000	1.000				X
SUPINE 2	0.994	1.000	1.63			X
SUPINE 3	0.973	0.964	6.35	7.35	1.00	X
SUPINE 4	0.914	0.904	13.03	13.43	0.40	X
SUPINE 5	0.844	0.824	18.56	18.28	-0.28a	
SUPINE 6	0.823	0.784	19.59	21.13	1.54	
SUPINE 7	0.784	0.743	22.40	23.96	1.56	
SUPINE 8	0.666	0.703	27.85	25.78	-2.07a	
SUPINE 9	0.594	0.555	31.79	34.18	2.39	

Continues on next page

Appendix I: Canadian (original) and Dutch item location of the individual items of the AIMS ***(Continued)***

Item	Proportion of infants passing the item		Item location Age (wks)			Item not used for equating
SIT1	0.991	0.976		4.66		X
SIT2	0.897	0.798	13.98	20.44	6.46	
SIT3	0.873	0.844	16.35	17.00	0.65	
SIT4	0.799	0.647	21.39	29.44	8.05	
SIT5	0.780	0.609	22.89	30.90	8.01	
SIT6	0.725	0.555	25.79	33.99	8.20	
SIT7	0.681	0.515	27.84	35.96	8.12	
SIT8	0.636	0.491	29.94	37.27	7.33	
SIT9	0.592	0.437	31.92	40.02	8.10	
SIT 10	0.435	0.441	39.74	39.67	-0.07a	
SIT 11	0.490	0.399	36.59	42.91	6.32	
SIT 12	0.400	0.349	40.95	45.38	4.43	
STAND 1#	0.996	1.000				X
STAND 2	0.962	0.926	3.74	10.18	6.44	X
STAND 3	0.762	0.493	22.93	36.91	13.98	
STAND 4	0.470	0.377	37.53	44.65	7.12	
STAND 5	0.450	0.361	38.47	45.51	7.04	
STAND 6	0.449	0.301	38.50	48.68	10.18	
STAND 7	0.350	0.303	44.45	49.35	4.90	
STAND 8	0.428	0.317	39.54	47.04	7.50	
STAND 9	0.381	0.293	41.99	50.33	8.34	
STAND 10	0.355	0.232	43.45	53.83	10.38	
STAND 11	0.281	0.160	47.65	59.69	12.04	
STAND 12	0.229	0.112	50.99	65.41	14.42	
STAND 13	0.204	0.108	52.70	67.05	14.35	
STAND 14	0.176	0.096	55.91	67.50	11.49	X
STAND 15	0.191	0.088	53.89	67.63	13.74	X
STAND 16	0.167	0.080	56.19	69.00	12.81	X

#=Regression estimates could not be calculated and suggest that more than 50% of infants are able to accomplish this item at birth.
a=The age of 50%
of the infants passed these items was younger in the contemporary data set compared to the original data set.

Part I

Infant and environmental factors

Chapter 3

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

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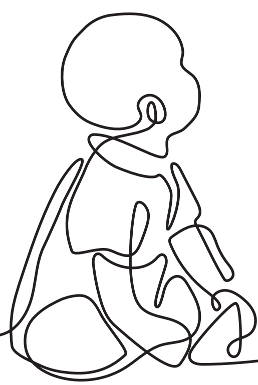

Abstract

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.

Keywords: child and environmental factors, cohort studies, gross motor development, infant, longitudinal design, systematic review

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 (2007), 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 [7]. 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 I**. 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. (2019) [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 (2019) [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 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 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 **(Appendix II: Table B1 and B2)**.

Table 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 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 71,546 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 **(Appendix II, Table B1 and B2)** 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 2**. 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. A summary of the significant associations of factors with GMD is displayed in **Table 3**.

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 **(Table 4)**.

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 (2018) 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].

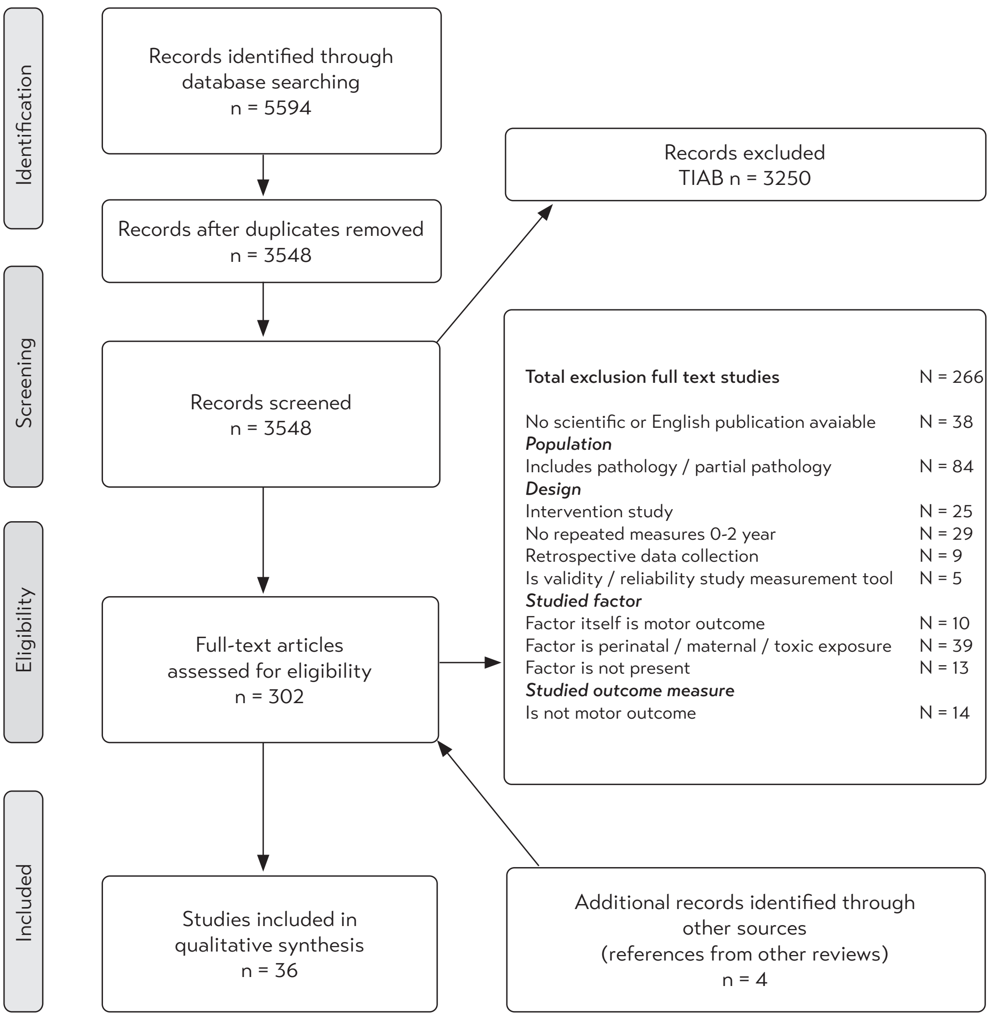

Figure 1: *PRISMA flowchart*

Table 2: Included studies and factors examined

Single factor studies

	Child factors						Environmental factors										
Author (year)	**GA**	**BW**	**Twin**	**Antropometry**	**Background**	**Motivation to move**	**Sleep Position**	**BF**	**Mat. Depression**	**Maternal Mental health**	**Adole -scent mother**	**Parental Mental health**	**Parental Neonatal Perceptions**	**Baby Walker**	**Day-care Attendance**	**Season of birth**	**Cultural context**
Yaari (2018)	Xc																
Espel (2014)	Xc																
Field (1978)	X																
Datar (2009)		Xc															
Gratham-McGregor (1998)		Xc															
Lung (2009)		Xc															
Nan (2013)			Xc														
Brouwer (2006)			Xc														
Goetghebuer (2003)			Xc														
Wilson (1975)			Xc														
Scharf (2016)				Xc													
Slining (2010)				Xc													
Bartlett (1998)				X													
Capute (1985)					Xc												

Atun-Einy (2013)						X											
Majnemer (2006)							Xc										
Davis 1998)							Xc										
Lung (2011)							Xc										
Ratliff (2001)							Xc										
Jardi (2017)								Xc									
Michels (2017)								Xc									
Morris (1999)								Xc									
Oddy (2011)								Xc									
Smith-Nielsen (2016)									Xc								
Sutter-Dallay (2011)									Xc								
Lung (2011)										Xc							
de Borba (2015)											X						
Lung (2009)												Xc					
Hernandez(2011)													Xc				
Siegel (1999)														Xc			
Souza (2010)															X		
Tsuchiya (2012)																Xc	
Vierhaus (2011)																	X

Continues on next page

3

Table 2: Included studies and factors examined **(Continued)**

Multiple factor studies													
	Child factors						Environmental factors						
Author (year)	GA	BW	Sex	Weight increase	Larger head	Score on cognition	Days of mechanical ventilationn	BF	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

Xc = Considered confounders ⟶ considered and final model confounders are represented in **Appendix III**

Table 3: Associations between factors and infant GMD

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***						

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Table 3: Associations between factors and infant GMD **(Continued)**

	RoB	Age of motor assessment outcome					Age of motor milestone outcome				
		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 * / **/ **/ **		

Environmental factors

	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/				
sLBW/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		sm*		
Parental neonatal perception							
Hernandez, 2011	H		sm*	sp**			
Babywalker							
Siegel, 1999	H		s ***			s***/ **	s**
Daycare attendance							
Souza, 2010	H			s	s		

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Table 3: Associations between factors and infant GMD **(Continued)**

		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
Season of birth winter											
Tsuchiya, 2012	H		s***	s***	ns						
Cameroonian vs German culture											
Vierhaus, 2011	H		s ns								

Multiple factors

Bjarnadottir, 2019	H						ns	ns	ns	ns	
Pereira, 2016	H		s								
Flensborg, 2017	H						s***	s***	s***		

*Note: = 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*

Table 4: Methodological Quality Assessment (QUIPS) (Hayden, 2019)

Child factors

	Study Participation	Study Attrition	Prognostic Factor Measurement	Outcome Measurement	Study Confounding	Statistical analysis and Reporting	Overall RoB
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 circum-ference							
Scharf, 2018	?	?	+	+	+	?	H
Overweight							
Slining, 2010	+	+	+	+	+	+	L

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Table 4: Methodological Quality Assessment (QUIPS) (Hayden, 2019) **(Continued)**

	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

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
Babywalker							
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

3

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. (2017) showed that birthweight in addition to GA explained most of the variance in motor milestone attainment [17]. 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 (2006) 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. reported that twins from 0 to 12 months scored lower on GMD, compared with singletons [27]. These outcomes are broadly in line with the study by Goetghebuer et al. (2003). After adjusting for the confounder birth-weight, 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. (1975) 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. (2006), prone-sleeping infants showed better GMD at 6 months, but not at 4 and 15 months. Davis et al. (1998) 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'.

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. (2011), supine-sleeping infants showed a delay in GMD at 6 months: at 18 and 36 months, the association was no longer present [33]. Ratliff et al. (2001) studied a population of very preterm (VPT) infants. GMD at the corrected ages of 4 and 13 months was not associated with sleeping supine [34].

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. 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 [35]. 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. (1999), 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. (2017) did not find an association of exclusive breastfeeding and infant GMD, nor for preterm infants [36]. The study by Oddy et al. (2011) 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. (2019) 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 (2016), 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. (2011) 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. (2009) who reported a significant negative association between the GA of VPT children and GMD [4]. The study of Espel et al. (2014) 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. 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 (2002), who found delays in GMD in both full-term and preterm infants with LBW [50]. These outcomes show that the impact of birthweight on GMD transcends that of premature birth. Golding et al. (2014) 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 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. (2003) 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. (2006), no significant differences were observed in GMD between twins and singletons with normal birthweight and GA. Unlike the study of Goetghebuer et al.(2003), who used the mean age of reaching a milestone, Brouwer et al. (2006) used the percentage of twins who achieved a milestone at a fixed age, which is less accurate and might explain differences in outcomes [25,26]. 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.

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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. (2014) which included six cross-sectional studies and also found no clear evidence for any association of breastfeeding with GMD. Despite these unequivocal findings, Jardi et al. (2019) 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 breast-feeding 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.(2007) 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. (2011) concluded that supine sleepers only showed a delayed GMD at 6 months, not at 18 and 36 months [33]. 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 Ayogi et al. (2009), including the study of Smith-Nielsen, also found no association between GMD and PPMD [58]. The studies of Smith-Nielsen et al. (2016) and Sutter-Dallay et al. (2011) 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 (1999), included in this review, was included in both reviews. Pin et al. (2007) reported conflicting evidence; Burrows and Griffiths (2002) 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 (1999). 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. (2013) 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.

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.*

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Appendix I: Search strings databases

Searchstring MedLine (PubMed) Run: Feb 2016 to Feb 2020 ((motor development*[tiab] OR "Motor Skills"[Mesh] OR motor skill*[tiab] OR motor milestone*[tiab] OR motor assessment*[tiab] OR motor behavior*[tiab] OR motor abilit*[tiab] OR motor growth[tiab] OR motor maturation[tiab] OR neuro maturation[tiab]) AND ("Child"[Mesh] OR "Infant"[Mesh] OR child*[tiab] OR newborn*[tiab] OR preschool*[tiab] OR infant*[tiab] OR neonate*[tiab]) AND (factor*[tiab] OR affordance* [tiab] OR constraint*[tiab] OR obstacle*[tiab] OR impediment*[tiab] OR enabler*[tiab] OR motivat*[tiab] OR inhibit*[tiab] OR stimulat*[tiab] OR correlat*[tiab] OR determin*[tiab] OR facilitat*[tiab] OR barrie*[tiab]) AND ("Cohort Studies"[Mesh] OR cohort stud*[tiab] OR concurrent stud* [tiab] OR cohort analys*[tiab] OR incidence stud*[-tiab] OR cohort survey*[tiab] OR follow up stud*[tiab] OR followup stud*[tiab] OR follow up analys*[tiab] OR followup analys*[tiab] OR follow up survey*[tiab] OR followup survey*[tiab] OR longitudinal stud*[tiab] OR longitudinal analys* [tiab] OR longitudinal survey*[tiab] OR prospective stud*[tiab] OR prospective analys*[tiab] OR prospective survey*[tiab] OR retrospective stud* [tiab] OR retrospective analys*[tiab] OR retrospective survey*[tiab] OR repeated measure*[tiab])) AND ("2001/01/01"[PDat]: "3000/12/31"[PDat])

Searchstring CINAHL (EBSCO) Run: Feb 2016 to Feb 2020 ("motor development*" OR MH "Motor Skills+" OR "motor skill*" OR "motor milestone*" OR "motor assessment*" OR "motor behavior*" OR "motor abilit*" OR "motor growth" OR "motor maturation" OR "neuro maturation") AND (MH "Child+" OR MH "Infant+" OR child* OR newborn* OR preschool* OR infant* OR neonate*) AND (factor* OR affordance* OR constraint* OR obstacle* OR impediment* OR enabler* OR motivat* OR inhibit* OR stimulat* OR correlat* OR determin* OR facilitat* OR barrie*) AND (MH "Prospective Studies+" OR "cohort stud*" OR "concurrent stud*" OR "cohort analys*" OR "incidence stud*" OR "cohort survey*" OR "follow up stud*" OR "followup stud*" OR "follow up analys*" OR "followup analys*" OR "follow up survey*" OR "followup survey*" OR "longitudinal stud*" OR "longitudinal analys*" OR "longitudinal survey*" OR "prospective stud*" OR "prospective analys*" OR "prospective survey*" OR "retrospective stud*" OR "retrospective analys*" OR "retrospective survey*" OR MH "Repeated Measures" OR "repeated measure*") Limiters: Publication year 2001-

Searchstring PsycInfo (EBSCO) Run: Feb 2016 to Feb 2020 (DE "Motor Development" OR DE "Perceptual Motor Development" OR DE "Psychomotor Development" OR "motor development*" OR DE "Motor Skills" OR "motor skill*" OR. "motor milestone*" OR "motor assessment*" OR "motor behavior*" OR "motor abilit*" OR "motor growth" OR "motor maturation" OR "neuro maturation") AND (child* OR newborn* OR preschool* OR infant* OR neonate*) AND (factor* OR affordance* OR constraint* OR obstacle* OR impediment* OR enabler* OR motivat* OR inhibit* OR stimulat* OR correlat* OR determin* OR facilitat* OR barrie*) AND (ZC "prospective study" OR ZC "retrospective study" OR ZC "followup study" OR ZC "longitudinal study" OR "cohort stud*" OR "concurrent stud*" OR "cohort analys*" OR "incidence stud*" OR "cohort survey*" OR "follow up stud*" OR "followup stud*" OR "follow up analys*" OR "followup analys*" OR "follow up survey*" OR "followup survey*" OR "longitudinal stud*" OR "longitudinal analys*" OR "longitudinal survey*" OR "prospective stud*" OR "prospective analys*" OR "prospective survey*" OR "retrospective stud*" OR "retrospective analys*" OR "retrospective survey*" OR MH "Repeated Measures" OR "repeated measure*") Limiters: Publication year 2001-

Searchstring SCOPUS (Elsevier) Run: Feb 2016 to Feb 2020 TITLE-ABS-KEY ("motor development" OR "motor skill" OR "motor milestone" OR "motor assessment" OR "motor behavior" OR "motor ability" OR "motor growth" OR "motor maturation" OR "neuro maturation") AND TITLE-ABS-KEY("infant" OR "child" OR "newborn" OR "neonate" OR "preschool") AND TITLE-ABS-KEY("factor" OR "affordance" OR "constraint" OR "obstacle" OR "impediment" OR "enabler" OR "motivate" OR "inhibit" OR "stimulate" OR "correlate" OR "determiner" OR "facilitate" OR "barrier") AND TITLE-ABS-KEY("cohort study" OR "concurrent study" OR "cohort analysis" OR "incidence study" OR "cohort survey" OR "follow up study" OR "followup study" OR "follow up analysis" OR "followup analysis" OR "follow up survey" OR "followup survey" OR "longitudinal study" OR "longitudinal analysis" OR "longitudinal survey" OR "prospective study" OR "prospective analysis" OR "prospective survey" OR "retrospective study" OR "retrospective analysis" OR "retrospective survey" OR "repeated measure") AND (PUBYEAR > 2000)

Appendix II: Table B1 Table with 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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Appendix II: Table B1 Table with characteristics and results of the included studies **(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 - = 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 = 20,112 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 value = 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.

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Appendix II : Table B1 Table with characteristics and results of the included studies **(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
Brouwer (2006, The Nether-lands)	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 ($p < 0.001$).
Goet-ghebuer (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 SDs 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]).

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Appendix II: Table B1 Table with characteristics and results of the included studies **(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
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.

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 ($p = 0.06$).

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Appendix II : Table B1 Table with characteristics and results of the included studies **(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
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 assess-ment* 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 ($r2 = 0.27$, $p = 0.0001$) and the total raw score were predicted by sleep position (prone versus supine) ($r2 = 0.17$, $p = 0.0001$), 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 ($r2 = 0.8$, $p = 0.048$). At 15 months of age, prone sleepers attained motor milestones significantly earlier: walking upstairs ($p = 0.04$) and walking ($p = 0.05$)

Davis (1998, USA)	n = 351 M BW = 3490 g (SD = 41) M GA not reported	Motor: MM 9 motor mile-stones Age: 0 - 18 months	Factor: sleep position Measurement: position recorded by parents: prone and supine Age easurement: 2-6 months	BW* Sex* Maternal education* Ethnicity* Number of siblings*	Linear regression analysis shows that prone sleepers acquire motor mile-stones significantly earlier for: rolling prone to supine ($p < 0.002$), sitting unsupported ($p = 0.003$), creeping ($p = 0.0002$), crawling ($p = 0.003$) and pull-ing to stand ($p = 0.001$). Walking alone was not associated with prone sleeping ($p = 0.4$). Increased prone playtime was associ-ated with tripod sitting, sitting alone, crawling and pulling to stand ($p < 0.05$). After controlling for maternal educa-tion, ethnicity, sex, BW and number of siblings, only pulling to stand remained significant ($p < 0.01$).
Lung (2011, Tai-wan)	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.

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Appendix II : Table B1 Table with characteristics and results of the included studies **(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
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).

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 morbiity*	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).

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Appendix II : Table B1 Table with characteristics and results of the included studies **(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
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-Dal-lay (2011, France)	n = 515 BW < 2500g = <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.
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).

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Appendix II : Table B1 Table with characteristics and results of the included studies **(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 (2009, Taiwan)	n = 17,595 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	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.

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Appendix II: Table B1 Table with characteristics and results of the included studies **(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; $p < 0.001$; -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; -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	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 ≤ 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).

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Appendix II : Table B1 Table with characteristics and results of the included studies **(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)	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.

Appendix II : Table B2 Table with confounders of the included studies

Author (year)	Child confounders									Environmental confouders								
	Age	Anthropometry	Birth order	BW	Ethnicity	GA	Plurality	Sex	Other child factors	Marital status	Maternal age	Maternal edu-cation	Num-ber 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														

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Appendix II : Table B2 *Table with confounders of the included studies* **(Continued)**

Author (year)	Child confounders									Environmental confouders								
	Age	Anthropometry	Birth order	BW	Eth-nicity	GA	Plurality	Sex	Other child factors	Marital status	Maternal age	Maternal edu-cation	Num-ber 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 back-ground																		
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}
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	

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Appendix II : Table B2 *Table with confounders of the included studies* **(Continued)**

Parental neonatal perception																		
Hernandez (2011)				-		X					-							$X_{31,32}$
Babywalker																		
Siegel (1999)												X					X	
Daycare attendance																		
Souza (2010)																		
Season of birth																		
Tsuchiya (2012)				-		-		-			-			-	$-_s$	-		
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-independance 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)

PART II

Infants

Chapter 4

Part II

Infants

Chapter 4

Modeling gross motor developmental curves of extremely and very preterm infants using the AIMS home-video method

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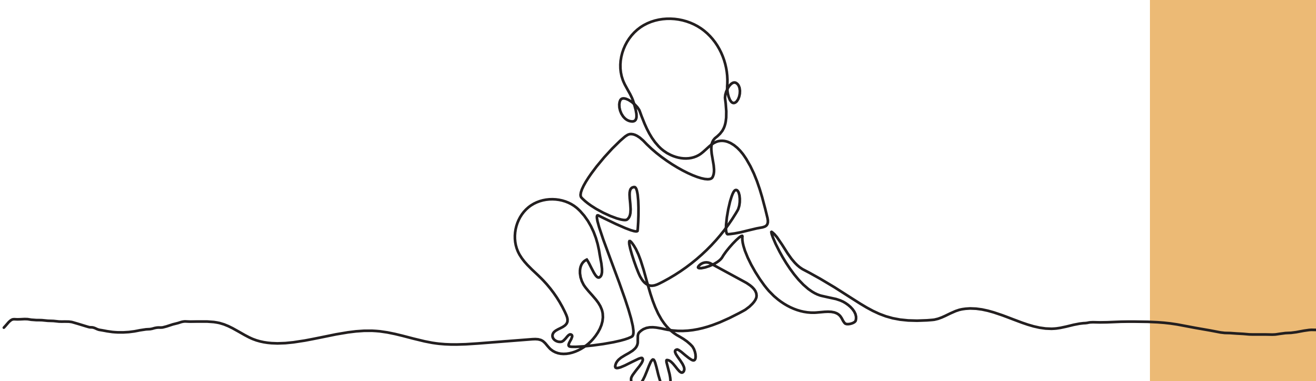

Abstract

Background: Motor development is one of the first signals to identify whether an infant is developing well. For very preterm (VPT) infants without severe perinatal complications, little is known about their motor developmental curves.

Aims: Explore gross motor developmental curves from 3 until 18 months corrected age (CA) of VPT infants, and related factors. Explore whether separate profiles can be distinguished and compare these to profiles of Dutch term-born infants.

Study design: Prospective cohort study with parents repeatedly recording their infant, using the Alberta Infant Motor Scale (AIMS) home-video method, from 3 to 18 months CA.

Subjects: Forty-two Dutch infants born 32.0 weeks gestational age and/or with a birthweight (BW) of <1500 grams without severe perinatal complications.
Outcome measures: Gross motor development measured with the AIMS.

Results: In total 208 assessments were analyzed, with 27 infants ≥ five assessments, 12 with < four, and three with one assessment. Sigmoid-shaped gross motor curves show unidirectional growth and variability. No infant or parental factors significantly influenced motor development, although a trend was seen for the model where lower BW, five-minute Apgar score <7, and Dutch native-speaking parents were associated with slower motor development. Three motor developmental profiles of VPT infants were identified, early developers, gradual developers, and late bloomers, which until 12 months CA are comparable in shape and speed to profiles of Dutch term-born infants.

Conclusions: VPT infants show great intra- and interindividual variability in gross motor development, with three motor profiles being distinguished. From 12 months CA onwards, VPT infants appear to develop at a slower pace. With some caution, classifying infants into motor developmental profiles may assist clinical decision-making.

Keywords: premature infants, motor development, Alberta Infant Motor Scale, developmental curves, longitudinal study

Introduction

Improved care of preterm infants has influenced clinical decision-making tremendously over recent decades [1]. From the moment an infant is expected to be born very premature (VPT) (before 32 weeks gestational age (GA)), clinical decisions are constantly made to support survival and developmental outcomes of the infant at risk [2]. VPT infants are at risk of developmental problems and identifying these problems makes early interventions possible [3-5]. Motor development is one of the markers to identify whether an infant is developing well and as such is one of the first signals to support clinical decision-making regarding starting early intervention [4,6]. However, the course of motor development for both preterm and term-born infants is known to be variable and non-linear, which makes predicting infant motor development and clinical decision-making difficult [7-10].

Research into factors affecting the course of motor development supports decision-making in follow-up, shaping early interventions, and decreasing parental concerns [11,12]. For premature infants, factors identified with sufficient evidence of a longitudinal association with gross motor development are mainly perinatal ones [4,13]. Only for the infant factors birthweight (BW) and gestational age (GA) is there strong evidence of a longitudinal association of lower BW and/or lower GA with more delayed gross motor development [5,14-20]. Few social, environmental, and parental factors, like parental education and socioeconomic status, have been subjects of longitudinal studies with premature infants. Further, research on these parental factors mainly concerns those associated with cognitive, and not motor, development [4].

Clustering of data, which enables the creation of profiles based on similarities and differences [21-23], can also contribute to clinical decision-making. Recent research into gross motor curves of typically developing Dutch term-born infants from 3 to 15 months, measured with the Alberta Infant Motor Scale (AIMS), has demonstrated that by using cluster analysis three profiles can be distinguished, based on different gross motor curves. The profiles are termed early developers, gradual developers, and late bloomers [10].

For premature infants, few studies examined distinct profiles in motor developmental trajectories. Most of these studies, mainly cross-sectional, group premature infants according to their GA: extremely premature (<28 weeks GA), very premature (28-32 weeks GA), and moderate-to-late premature infants (32-37 weeks GA) [24]. Their results support the idea that preterm infants present with lower scores on motor performance, even when corrected for prematurity, than their term-born peers [16]. Other studies with a longitudinal design mainly determine the shape of the motor developmental trajectory with the focus on the (in)stability of longitudinal measurements of VPT infants at risk. For instance, Janssen et al. (2011) reported three profiles in gross motor development measured with the Bayley Scales of Infant Development, 2nd edition (BSID-II) Motor Scale among a sample of 348 preterm infants (≤32 weeks GA) at 6, 12, and 24 months CA. They described these clusters in terms of the (in)stability of the motor trajectory: stable, relatively stable, and unstable classifications [7]. Erikson et al. (2003) reported two profiles for a sample of 165 very low birthweight (VLBW) infants followed over a wider age span, namely from 5 months (CA) to 5.5 years, measured with the Movement Assessment of Infants (MAI): stable or unstable motor development [25]. Lastly, Su et al. (2017) assessed preterm infants with VLBW four times, at 4, 6, 9, and 12 months, with the AIMS, finding three distinct motor profiles: stably normal (53%), deteriorating (32%), and persistently delayed (13%) [26]. These three studies included very premature infants with and without perinatal complications, such as severe intraventricular hemorrhage (IVH), bronchopulmonary dysplasia (BPD), and brain damage [7,25,26].

Little is known about the motor developmental curves of VPT infants without severe perinatal complications. The focus of the present study was on the shape and speed of individual gross motor developmental curves from 3 to 18 months CA in Dutch infants born VPT and/or weighing less than 1500 grams. Within the sample, we explored which factors were related to the course of gross motor curves. Lastly, we examined whether profiles of gross motor development could be distinguished, and how they related to such profiles for a sample of term-born (TB) Dutch infants. The results of this study may contribute to clinical decision-making, shaping early interventions, and informing realistic parental expectations.

Methods

Participants and procedure

In this prospective cohort study, the GODIVA-PIT study (Gross mOtor Development of Infants using home-Video registration with the Aims - following Premature Infants in Time), infant gross motor development was assessed at seven time-points between 3 and 18 months CA. The age of 18 months was chosen to ensure to the inclusion of VPT infants who had reached the milestone of independent walking [27].

In the Netherlands, motor development of premature infants is monitored according to the European Standards of neonatal follow-up. Infants visit the neonatal follow-up clinics spread across the country at the ages of 6, 12, and 24 months CA, and at 5 and 8 years. At 6 and 12 months CA, the AIMS is administered to assess gross motor development [28]. After discharge from the hospital, infants born before 32 weeks GA and/or with a BW less than 1500 grams are advised to participate in the TOP program (Transmural developmental support for VPT infants and their parents), a post-discharge responsive parenting program for VPT infants and their parents performed at home by trained pediatric physical therapists, covered by the Dutch health insurance system [29].

Parents were recruited between May 2017 and December 2019 in the Wilhelmina Children's Hospital (Utrecht), Radboud University Medical Centre (Nijmegen), Isala Hospital (Zwolle), and by TOP pediatric physical therapists throughout the Netherlands. Recruitment took place at the regular neonatal, outpatient follow-up, or during parents' first contact with the TOP therapist.

Infants born before or at 32 weeks GA and/or with a BW of <1500 grams who at the start of the study were younger than 7 months CA were eligible for the study. Due to changes in the follow-up protocol at the outpatient clinic of the Wilhelmina Children's Hospital in January 2018 whereby all infants were no longer seen at their term date, and as their first visit was at 6 months, the inclusion criteria were broadened to 7 months of age. Their parents had to understand Dutch language.

Exclusion criteria were: a known syndrome, a neuromuscular disorder, severe neuroimaging abnormalities (e.g., cystic periventricular leukomalacia, IVH Grade III or IV), meningitis, bronchopulmonary dysplasia (defined as oxygen supplementation >36 weeks postmenstrual age), congenital anomalies, necrotizing enterocolitis requiring surgical procedures, prolonged tube feeding (defined as beyond hospital discharge), and severe visual or hearing disorder.

When infants met the inclusion criteria, parents were invited to participate in the study and sent information, accompanied by informed consent forms. After approximately a week, parents were contacted and asked if they intended to participate and/or had any questions. If they agreed to participate, parents were asked to return the signed informed consent forms and booklet with information, checklists [30], and instructions were sent to them.

Data for Dutch term-born infants were used retrospectively. These data originated from the GODIVA-KIT (children following in time) study, a prospective longitudinal study with the objective of modeling motor growth and exploring different patterns in gross motor trajectories in a sample of term-born (>37 weeks GA) typically developing Dutch infants from 3 to 15 months, using AIMS raw scores. Data for the GODIVA-KIT study were collected between 2016 to 2018. The present study had a similar protocol but a different population than the GODIVA-KIT study [10].

AIMS home-video method

Parents used the AIMS home-video method to collect data. This method is validated and reliable for assessing the AIMS [31,32]. Parents received instructions, comprising three instruction videos and a booklet with three corresponding checklists on how and what to record and how to upload their videos to a secure digital server. After uploading, the researcher/pediatric physical therapist (IS) assessed the video using the AIMS and sent parents feedback on their infants' motor development by email. Whenever abnormalities were seen in the infant's motor presentation, the attending physician and/or pediatric physiotherapist were contacted for consultation.

Parents had a window of two weeks to plan a time to record their infant. Before this window began, parents received a reminder by email of the actual dates of the recording window (also noted in the information booklet). An additional reminder was sent one week after the start of the window. Furthermore, parents received the Parental Beliefs questionnaire [33] accompanied by some demographic questions, before the first time of recording, and again when their infant was 15 months CA. Parents were asked to film their infant with the AIMS home-video method five to seven times, depending on the CA of their infant at the start. The interval between recordings was 2 or 3 months **(see Figure 1)**.

Measurement

The AIMS measures infant gross motor development from birth until independent walking. It is a norm-referenced observational instrument with good psychometric properties [34]. Most of the 58 items are based on spontaneous movements of the infant in four different postures (supine, prone, sitting, and standing). The actual motor repertoire of the infant is represented by the observed items.

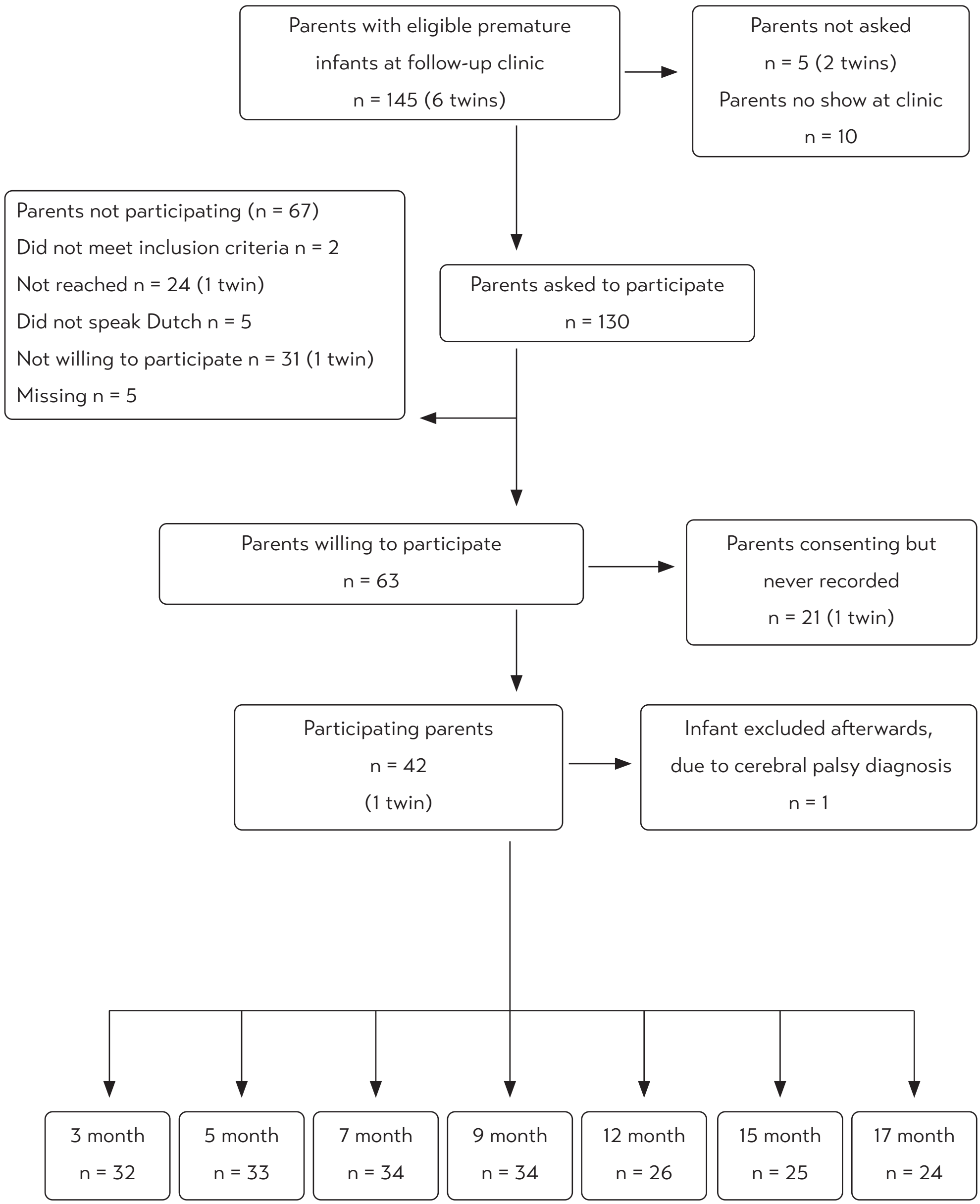

Figure 1: *Flow-chart eligible and participating infants*

Ethics

The GODIVA-PIT study was approved by the Medical Ethical Board of the University Medical Centre Utrecht (METC/UMCU) with protocol nr. 17-186/C. Parents gave written informed consent prior to participation. Video data were stored on a secure server at Utrecht University of Applied Sciences.

Data analysis

Population characteristics were calculated with frequencies, percentages, means, standard deviations (SD), and ranges. Next to the descriptive analysis, characteristics of participating infants and parents, drop-outs and non-participators were compared with a one-way ANOVA. Next, three different components were analyzed:

1. Trajectories of gross motor development and associated factors,
2. Modeling Dutch preterm gross motor profiles
3. Comparison between Dutch term-born and preterm gross motor profiles.

Trajectories of gross motor development and associated factors

Linear Mixed Model analysis (LMM) was used, suitable for hierarchical data structures such as repeated measures over time. LMM allows observations to be interdependent, and every observation is considered a data point [35,36]. Hence, LMM analysis is considered most applicable for a relatively small sample with missing data points.

The first step in LMM is to explore which model (linear, quadratic, or cubic) fits best. The intercept and slope were allowed to vary across individuals. Therefore, age is considered a random factor with a random intercept because exploration of the gross motor trajectories shows that each infant has a different start and course in motor development. The best fit model is determined by the Akaike information criterion (AIC), which needs to decrease to be a better fit [37].

The second step was LMM with backward selection of factors representing infant and parent characteristics. The characteristics BW, GA, sex, five-minute Apgar score, maternal age, paternal age, maternal education, paternal education, birth order, and parental mother tongue (i.e., one or both have mother tongue Dutch) were considered as potential factors influencing gross motor development. BW was divided into three categories: <1000 grams, 1000-1499 grams, and ≥1500 grams. The five-minute Apgar score (Apgar5) comprised two categories: <7 or ≥7. Maternal and paternal age were divided into categories starting from 25 years in steps of 5 years to 40 years of age. Maternal and paternal education had the categories: primary, secondary lower, secondary higher, and tertiary. These factors were assumed to be fixed because they do not change over time. For the factors sex and BW, an interaction effect with age was added. The factors were sequentially deleted, based on the highest p-value and $p > 0.05$, from the models to finish with the model that explains the data best. The AIC was decisive in determining the best model with a minimum difference of 3. The factors GA and BW were highly correlated ($r = 0.631$, $p < 0.001$), and also the factors maternal and paternal age were highly correlated ($r = 0.729$, $p = 0.001$). To avoid multicollinearity, GA and paternal age were left out of the analysis. AIMS raw scores were used as the outcome variable because these are not norm-referenced [7].

Modeling Dutch preterm gross motor profiles

We explored whether gross motor profiles similar to those of Dutch term-born infants (i.e., early developer, gradual developer, and late bloomer) could be identified for the preterm infants. A hierarchical cluster analysis, first with a dendrogram and subsequently with K-means cluster analyses, was performed to confirm whether three profiles were a possibility, and infants were assigned to the initial calculated profile. The mean age and standard deviation of independent walking of each profile were calculated. For infants with one or two assessments, the initial calculated profile was compared to the profile based on the age of independent walking ± 1SD. An infant was reassigned when the age of independent walking fitted a different profile.

When the age of independent walking was not available, the classification according to the initial analysis was preserved. With ANOVA's tests, the profiles were tested on their differences. Because of the small number of infants in a profile, descriptive analyses were used to gain more insight into the characteristics of the three profiles.

Comparison between Dutch term-born and preterm infant gross motor profiles

To compare the profiles of the Dutch term-born (TB) infants (TB early developer, TB gradual developer, TB late bloomer) with the Dutch premature sample (VPT early developer, VPT gradual developer, VPT late bloomer), at all different ages, a two-way ANOVA was performed to analyze the interactions between the group (Term (TB) or Preterm (VPT)) and developmental profile (early developer, gradual developer or late bloomer).

IBM SPSS statistics package for Windows, Version 25.0. was used for statistical analyses.

Results

Figure 1 shows the flow chart of all infants assessed. A total of 145 infants were eligible, of whom 43, with their parents, participated. Because one infant was diagnosed with cerebral palsy during follow-up, the data of 42 infants were used for analyses. Reasons for not participating were: no show at follow-up, parents not approached, not willing to participate, not speaking Dutch, or not reached after the first contact.

The mean BW was 1205 (±330) grams and infants were born with a mean GA of 29.1 (±2.1) weeks. Boys and girls were equally distributed. 27 infants were assessed at least five times, 12 two to four times and three once. In total, there were 208 assessments (mean times filmed = 4.9).
Characteristics of infants and parents are displayed in **Table 1.**

There were no differences in the infant characteristics (sex, GA, BW, Apgar5, type of delivery) of the infants that dropped out or did not start the study.

Trajectories of gross motor development
Individual motor trajectories of the infants are presented in **Figure 2**. All infants show unidirectional growth and a sigmoid-shaped curve. A great deal of variety in acceleration and deceleration is seen at different times, which implies intra- and interindividual variation in gross motor curves. The biggest difference score (AIMS raw score), the mean number of items scored per month, is seen between 5 and 9 months CA (mean diff/months = 4.4 items; range 0 - 12.5 items), visible in **Figure 2** where the biggest acceleration between 5 and 9 months CA is evident.

The first step in LMM was to fit the best model based on the AIC (AIC= 1208), which was a cubic polynomial **(Appendix I)**. The second step was backward selection of the infant and parental factors. There was a trend that the model with the best fit (AIC = 1020) included the factors BW ($\beta 1$ = -4.10, $\beta 2$ =0 .004; p = 0.031), Apgar5 (β = -3.54; p = 0.033) and parental mother tongue (β= -3.16; p = 0.059) **(Table 2)**. This means that there is a trend that infants with lower BW, having a five-minute Apgar score <7 and having Dutch-speaking parents, are prone to lower AIMS scores. Leaving parental mother tongue out of the model, the factors BW and Apgar5 did not remain significant in the final model (AIC = 1021).

Table 1: Infant and parent characteristics according to developmental profile: early developer, gradual developer, and late bloomer

Infant characteristics		Total	Early developer (n = 10)	Gradual developer (n = 27)	Late bloomer (n = 5)
Sex	Male	21	5	13	3
	Female	21	5	14	2
Mean birthweight in grams		1205	1299	1195	1073
(±SD)		(±330)	(±319)	(±340)	(±304)
Birthweight category	Extremely low (<1000 gr)	11	1	8	2
	Very low (1000-1500 gr)	25	6	16	3
	Low (1500-2500 gr)	6	3	3	0
Mean gestational age in weeks		29.1	30.4	28.8	28.7
(±SD)		(±2.1)	(±1.3)	(±2.5)	(±1.3)
Gestational Age category	<28 wks GA	11	1	9	1
	28-30 wks GA	15	2	10	3
	≥30 wks GA	15	7	8	1
	>32 wks GA	1	-	1	-
Delivery	Vaginal delivery	20	4	14	2
	Caesarean section	21	6	12	3
	Not available	1	-	1	1
Five-minute Apgar score	<7	8	2	5	1
	≥7	32	8	20	4
	Not available	1	-	1	-
Age (CA) of independent walking	Mean age	15	12	15	19
(months)	(±SD)	(±2.8)	(±1.27)	(±1.47)	(±2.08)
	(Range)	(11-22)	(11-14)	(12-18)	(17-22)

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Table 1: Infant and parent characteristics according to developmental profile: early developer, gradual developer, and late bloomer **(Continued)**

Parent characteristics					
Maternal age (±SD)	(Range)	31.9 (±4.0)	30.2 (±3.1)	32.2 (±4.5)	33.6 (±0.9)
Paternal age (±SD)		34.4 (±3.6)	34.6 (±2.5)	34.1 (±4.2)	35.2 (±1.6)
Age category (maternal / paternal)	25-29 years	11 / 3	3 / 0	8 / 3	0 / 0
	30-34 years	22 / 15	7 / 4	11 / 9	4 / 2
	35-39 years	5 / 14	0 / 4	4 / 7	1 / 3
	40-45 years	3 / 1	0 / 0	3 / 1	0 / 0
	Not available	1 / 9	- / 2	1 / 7	- / -
Maternal / paternal education	No education	0 / 0	0	1 / 0	0 / 0
	Primary	1 / 0	0	0 / 0	0 / 0
	Secondary lower	0 / 3	0	0 / 3	0 / 0
	Secondary higher	5 / 9	1 / 2	4 / 7	0 / 0
	Tertiary	27 / 21	7 / 6	15 / 10	5 / 5
	Not available	9 / 9	2 / 2	7 / 7	- / -
Mother tongue	Dutch	34	7	22	5
	Other	7	3	4	0
	Not available	1	-	1	- / -

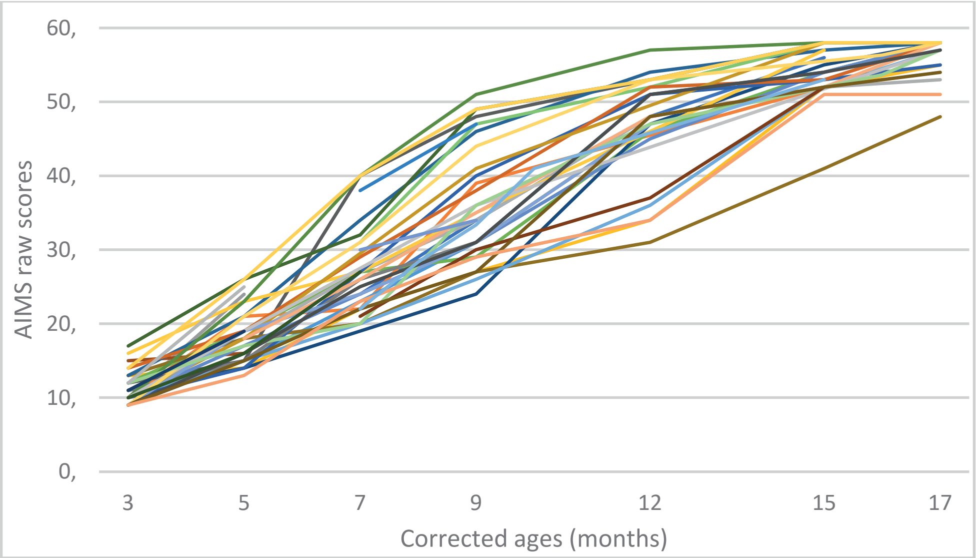

Figure 2 : *Individual gross motor developmental trajectories from 3 – 17 months CA in raw AIMS scores.*

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Table 2: Results of the Linear Mixed Model analysis of the model with the best fit.

	Estimate	SE	p	95% Confidence Interval	
Intercept	5.67	4.07	0.792	-2.36	13.70
Age	1.77	1.28	0.169	-0.76	4.29
Age*Age	0.33	.14	0.016	0.06	0.60
Age*Age*Age	-0.01	.00	0.001	-0.02	-0.01
Parental mother tongue	-3.16	1.62	0.059	-6.45	0.124
Apgar5	-3.54	1.58	0.033	-6.77	-0.31
BW <1000	-4.10	2.16	0.031	-8.48	0.28
≥1000	0.00	2.02		-4.09	4.09

Footnote:
For parental mother tongue, 'not Dutch' is the reference group
For five-minute Apgar score, ≥7 is the reference group
For BW, ≥1000 grams is the reference group

Modeling Dutch preterm gross motor profiles

Cluster analysis confirmed that it was possible to create three different profiles according to the dendrogram. Also, the two-step clustering revealed a good cluster quality when these three profiles were formed **(see Appendix II)**. K-means cluster analyses with three predefined clusters and excluding cases pairwise was necessary to assign each infant to a profile. In total, three infants, for whom only one or two measurements were available, were reassigned to another profile based on their age of reaching the milestone of independent walking. For the other seven infants with only one or two assessments, the age of independent walking was not available (n = 4) or the infant was correctly allocated in the class (n = 3). At all ages, the ANOVA showed significantly different AIMS raw scores ($p < 0.005$), except for 3 months CA ($p = 0.274$). At 3 months CA, the mean AIMS score for both early developers and late bloomers was 12 ($SD_{early\ developer}$ = ±2.7; $SD_{late\ bloomer}$ = ±2.1), for the gradual developers this was 11 (SD = ±2.0).

Preterm early developers (n = 10): For seven infants, the age of independent walking was known with a mean of 12 (±1.27) months CA (range 11-14). For the five infants with available assessments, all had a maximum score at the last assessment. Only one infant (out of four available assessments) had a score of 57 items, the other three already having the maximum AIMS score (58 items) at this age.

Preterm gradual developers (n = 27): For twenty infants, the age of independent walking was known, with a mean age of independent walking 15 (±1.47) months CA (range 12-18). Eight infants (out of 14 available assessments) achieved a maximum AIMS score at 17 months CA.

Preterm late bloomers (n = 5): For four infants, the age of independent walking was known, while, for one, it was known that he was not independently walking by his second birthday (20 months CA). The mean age of independent walking was 19 (±2.08) months CA (range 17-22), and none achieved all items on the assessment at 17 months CA.

Looking at the curves for the different profiles **(Figure 3)**, it is apparent that the early developers show a quadratic line, with a ceiling effect starting at 12 months CA. For the late bloomers and gradual developers, a more S-shaped curve (cubic line) is seen. Acceleration for the gradual developers starts at approximately 9 months CA and for the late bloomers at 12 months CA. The ceiling effect, deceleration in the curve, for the gradual developers starts at 12 months CA and for the late bloomers at the age of 15 months CA. The developmental curves of the different developmental profiles showed a significant effect of time (AIC 1150; p = 0.000) when added to the baseline model (AIC = 1208), which means that the three profiles differ in the pace of gross motor development. According to the ANOVA, there are no significant differences between infant and parental characteristics in the different profiles, probably due to the small number of late bloomers. Therefore, only descriptive statistics for each profile are presented **(Table 1)**.

Comparison between Dutch term-born and premature infant gross motor profiles

When combining data from the Dutch term-born (TB) and the premature infants (VPT) from this present study **(Figure 3)**, it is apparent that the shapes of the developmental curves are similar for all three profiles. There are some differences between the developmental pace of the profiles of TB gradual developers and late bloomers and VPT gradual developers and late bloomers. These differences become visible from 12 months CA onwards, where the interaction effect between group (TB or VPT) and profile (early developer, gradual developer or late bloomer) disappears. This implies that the differences in AIMS scores at different ages are explained by the effect of being preterm or term-born and being an early developer, a gradual developer, or a late bloomer. At 12 months (CA), there is a difference between the groups TB late bloomers and VPT late bloomers. There is also a difference in the profiles whereby the late bloomers differ from the early developers (mean differenceearly-late = -7.006, p = 0.001) and the gradual developers (mean differencegradual-late = -4.663, p = 0.007).

Scores at the 15 months (CA) assessment showed significant differences between the early developers and both the gradual developers and late bloomers (mean differencelate-early = -3.870, p = 0.010 and mean differencegradual-early = -2.694, p = 0.047).

In **Table 3,** the comparison of the mean AIMS raw scores of the term-born and preterm infants according to the profiles is shown. In addition, these scores are compared to the Canadian norm references to show which infants are at risk (score below -1SD) or have a motor developmental delay (score </= 5th percentile). The VPT late bloomers show a delay in their gross motor development from the age of 5 months, and the TB late bloomers are at risk of delay from 5 months. Also, the VPT gradual developers are at risk of developmental delay from 5 months (CA). The early developers in both groups (TB/VPT) do not show any (risk of) delay in their gross motor development at any ages.

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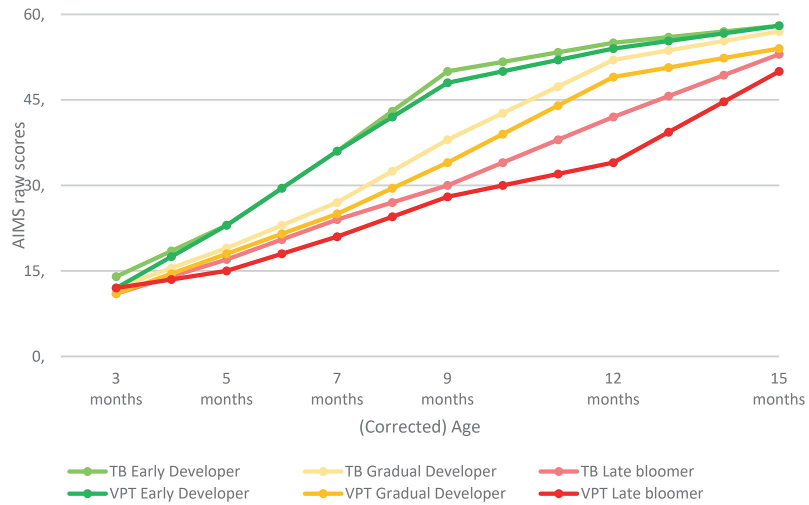

Figure 3: *Comparison of the three different gross motor curves, Early developer, Gradual developer, and Late bloomer, of the term-born (TB) infants and very preterm infants (VPT).*

Table 3: Comparison of the mean raw AIMS scores of the term-born and preterm infants, according to the profiles and the Canadian norms.

	Mean AIMS score (±SD)					
Age (CA)	TB Early Developer	PT Early Developer	TB Gradual Developer	PT Gradual Developer	TB Late Bloomer	PT Late B loomer
3 months	14 (±2.1)	12 (±2.7)	12 (±1.6)	11 (±2.0)	11 (±1.5)	12 (±2.1)
5 months	23 (±2.8)	23 (±3.9)	19 (±2.8)	*18* (±2.6)*	*17* (±3.2)*	**15** (±2.2)**
7 months	36 (±5.2)	36 (±4.0)	27 (±2.6)	*25* (±3.3)*	*24* (±2.9)*	**21** (±1.3)**
9 months	50 (±5.6)	48 (±2.1)	*38* (±4.2)*	*34* (±4.5)*	**30** (±2.6)**	**28** (±1.6)**
12 months	55 (±2.4)	54 (±1.6)	52 (±2.0)	*49* (±2.3)*	**42** (±5.3)**	**34** (±2.3)**
15 months	58 (±0.8)	58 (±0.5)	**57** (±1.7)**	**54** (±2.1)**	**53** (±2.9)**	**50** (±4.8)**
17 months		58 (±0.0)		**57** (±1.7)**		**53** (±4.0)**

**score below -1SD according to the Canadian norm references*
****score below 5th percentile / -2SD according to the Canadian norm references**

Discussion

This study explored the shape and speed of individual gross motor developmental curves from birth until 18 months CA in a sample of very premature (≤32 wks GA and /or <1500 grams BW) Dutch infants. Gross motor curves show unidirectional growth with a sigmoid shape, with interindividual variety. The biggest overall acceleration, as well as the largest variability between infants, was apparent between 5 and 9 months CA. In this sample, none of the infant or parental factors significantly influenced the shape and speed of motor development, with only a trend for the model with the factors BW, parental mother tongue, and five-minute Apgar score being seen. Cluster analysis distinguished three motor developmental profiles, namely early developers, gradual developers, and late bloomers. These profiles show significantly different developmental curves based on the total raw AIMS scores between 5 and 17 months CA. There were no significant differences between the developmental profiles regarding specific characteristics in these groups. Lastly, comparing the developmental profiles of our sample of VPT Dutch infants to those observed earlier in a sample of Dutch term-born infants, a similar shape of the curves was identified until 12 months CA. From 12 months CA onwards, the effect of being a premature infant is different for the gradual developers and for the late bloomers, where the preterm gradual developers and late bloomers appear to develop at a slower pace than the term-born infants, as seen in the delayed acceleration in the curves. For the early developers, there is no difference in the effect of being a term-born or preterm infant. Late bloomers were already showing a developmental delay (according to the Canadian norm references) from 5 months CA, whereas the early developers scored normal at all ages.

Shape of gross motor curves and factors of influence

Previous studies have reported similar findings concerning the course of gross motor development measured with the AIMS [6,10,38-40]. The acceleration in the curve was as expected since motor development in infancy is faster than at any other time during life [41]. Also, the large interindividual variability in the scores between 5 and 9 months is consistent with what is known about gross motor development as assessed with the AIMS. Furthermore, theories on infant development confirm the plausibility of typical infant development being characterized by variability [42,43].

Based on a previous systematic review of longitudinal studies [13], we explored the influence of several infant and parental factors on gross motor development. No factors of significant influence were found, which is probably due to the small sample size. The trend observed for the child factors BW and five-minute Apgar score to be associated with gross motor development is in line with previous findings. For example, BW is a well-researched factor with a profound and long-lasting influence on motor development, established in multiple longitudinal studies, with lower BW being associated with slower gross motor development [15,20,44]. In the present study, this was replicated in the model of best fit, which showed a lower BW to be associated with slower gross motor development. A five-minute Apgar score 7 indicates that the condition of the newborn is good to excellent and that the infant is adapting well to the environment [45,46]. A five-minute Apgar score 7 is associated with an increased risk of impaired neurodevelopmental outcomes, including motor [47]. A recent study examined the relationship between the five-minute Apgar score on the neurodevelopmental outcome of term-born infants from 8 to 66 months. Results showed that a five-minute Apgar score was inversely associated with neurodevelopmental delay [45]. This is in concordance with the results of our study, where, together with a lower BW and Dutch parental mother tongue, a lower five-minute Apgar score was associated with slower motor development.

Parental mother tongue in our study indicates parents who have a migration background, i.e., that at least one parent was born abroad (the first generation) or born in the Netherlands but whose parents were born abroad (second generation). These parents likely have different cultural backgrounds.

In several studies with the AIMS, cultural background appears to be a factor influencing motor developmental pace. In the Netherlands, infants' gross motor development seems to develop at a slower pace than Canadian and American infants measured with the AIMS [48,49] and Bayley Scales of Infant Development [50]. These cross-cultural differences in the pace of motor development are also observed in other populations [38,51,52]. Because of the small number of infants with non-Dutch native-speaking parent(s) included in our study, it is difficult to draw any conclusions but may explain why this factor approached significance ($p = 0.059$) in the model with BW and five-minute Apgar score.

Conflicting evidence exists in previous research regarding sex. Some studies reported differences between the development of (premature-born) boys and girls, with boys having more risk of developmental delay than girls [53]. In our study, no differences were found between boys and girls. This agrees with the study of Haastert et al. (2006) where 800 Dutch VPT infants at risk were measured with the AIMS. They reported that only at 7 to 8 months CA a difference was found between boys and girls, with girls scoring higher [38].

Profiles in gross motor curves

Comparing the results of our study with that of Su et al. in Taiwan where three profiles were also found, it is interesting to see that, at the ages of 9 and 12 months CA, the infants show similar mean AIMS raw scores [26]. Since in our study infants with no or only minor complications (IVH I-II), but without BPD and severe brain damage as in the study of Su et al., were included, it would be expected that the Dutch infants would have performed better. However, their sample consists of Taiwanese infants, and previous research seems to show, as stated earlier, Dutch infants develop at a slower pace than in other cultures [48,49, 50]

In our sample of VPT infants, with the same cultural context and methodology as that of the study of Dutch term-born infants by Boonzaaijer et al. [10], three motor developmental profiles were also identified, i.e., early developer, gradual developers, and late bloomers. The curves of these profiles appeared similar in shape to those of the term-born profiles. Surprisingly, the TB and VPT early developers also revealed significant similarities in the speed of their developmental curves. The VPT gradual developers and late bloomers show a decrease in gross motor developmental pace from 12 months CA compared with the TB gradual developers and late bloomers.

To our knowledge, there is little research available that confirms these specific results. The reasons may include that 1) the time frame of the measurements covers only the first year after birth, 2) larger age intervals are used between measurements, and 3) motor developmental analyses are performed on the entire sample, possibly resulting in higher average gross motor scores [7,16,25,26,38].

In a study by Wang et al. (2013) of Taiwanese VLBW infants, with and without PVL, compared to TB infants measured with the AIMS at 6, 12, and 18 months (CA), the former did not score significantly differently from the TB infants from 12 months onwards [54]. Although this study is not fully comparable to our study, it does provide information that there are VPT infants who develop similarly to TB infants.

In the study of Yaari et al. (2006), extremely preterm (n = 18), very preterm (n = 32), and moderately preterm (n = 53) infants were compared to full-term (n = 37) infants at multiple time-points, measured with the Mullen Scales of Early Learning (MSEL).

They showed that the TB infants increased in their motor score from 4 months onwards, whereas the EPT and VPT infants showed a decrease in their motor score from 4 to 8 months, an increase from 8 to 12 months, and a decrease from 12 to 18 months again [55]. Despite the difference in measurement instrument, with the Mullen scale being a composite score of which gross motor development is only a part, the results seem to support our findings that the VPT gradual developers and late bloomers decrease in gross motor developmental pace after 12 months CA. A possible explanation for this reduction in gross motor developmental pace is that difficulties in motor performance become gradually evident during the first years of life when more complex abilities start to emerge [56].

Limitations and strengths

The sample size of the present study is small, making it hard to draw firm conclusions. Moreover, due to the sample size, some analyses could not be performed. Nonetheless, LMM allows all assessments to be included in the analysis which made it possible to investigate gross motor curves and the factors influencing them.

Another limitation might be that most parents were highly educated, making the results unrepresentative of the whole population of parents with preterm infants in the Netherlands. Research indicates that lower socioeconomic status of parents may have a negative influence on motor development of the infant [57-60]. This may imply that our sample has performed better than can be expected of the general population of VPT infants.

Also, the generalizability of the results to the VPT population is not possible, because of the exclusion of infants having severe complications such as BPD, NEC, etc. These infants probably have a less favorable gross motor development, so with our results, one should take that into account [61, 62].

A strength of our study was that we gathered data at short age intervals during the first 18 months after birth. This made it possible to detect differences between VPT and TB infants at different ages which may help in decision-making and starting early interventions.

Clinical implications

Clinical decisions in neonatal follow-up are based on the information (concerns) parents provide, the results of standardized assessments, and the physician's observations [63], knowledge, and experience [64,65]. Altogether, neonatal follow-up not only aims to identify infants with severe gross motor impairments, like cerebral palsy but also to accurately identify infants with less severe gross motor impairments, who might likewise benefit from early intervention [63]. Distinguishing gross motor developmental profiles, combined with a knowledge of infant and parental factors, may help clinical decision-making about pediatric physiotherapy intervention.

Future research

For future research, it would be interesting to combine a larger data set of term-born infants with preterm-born infants to gain more insight into the range of possible gross motor developmental profiles. Perhaps more profiles will be distinguished, giving more direction for clinical decision-making and start interventions as early as possible. In addition, research should also focus on preterm infants who appear to develop at the same pace as term-born infants. For these early developers, less focus may be required for following their gross motor developmental domain, while still following them on other developmental domains. For the gross motor development follow-ups specific for these early developing infants, it may be a consideration to replace a ‘live’ assessment with a home-video consultation.

Gaining more insight into which factors explain the different profiles may also be of added value. To do so, we would recommend creating a larger and more representative sample, especially with regards to parental education and ethnicity, but also infants with more severe complications as is seen in clinical practice. We would also recommend considering research on VPT infants without complications at older ages, to gain a better understanding of infants with gross motor developmental problems and associated factors at preschool.

Lastly, with new and advanced technologies [66-68], it is perhaps possible to assess 'infants’ development more frequently. This will give us even more detailed information about infant motor development and perhaps an indication of periods when motor development is subject to change.

Conclusion

This study contributes to insights into gross motor development of VPT infants (<32 weeks GA and/or weighing <1500 grams) without severe perinatal complications, but still at risk in various other developmental domains. Distinguishing gross motor developmental profiles may contribute to clinical decision-making, shaping early interventions, and supporting realistic parental expectations. Future research should focus on clustering infants and possible explaining factors by assessing gross motor development more frequently.

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AUTHOR CONTRIBUTIONS

Imke Suir: Conceptualization, Methodology, Investigation, Resources, Data Curation, Formal Analysis, Writing – Original Draft, Project administration.
Marike Boonzaaijer: Conceptualization, Methodology, Investigation, Writing – Review & Editing.
Ora Oudgenoeg-Paz: Formal Analysis.
Paul Westers: Formal Analysis.
Linda de Vries: Resources, Validation, Writing - Review & Editing, Supervision.
Janjaap van der Net: Resources, Validation, Writing - Review & Editing, Supervision.
Jacqueline Nuysink: Conceptualization, Methodology, Writing - Review & Editing, Supervision.
Marian Jongmans: Conceptualization, Methodology, Writing - Review & Editing, Supervision.

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Appendix I: Cluster analysis

Appendix II: Comparison term and premature profiles (early developer, gradual developer, late bloomer)

PART III

Chapter 5

Chapter 6

Parents

Part III

Parents

Chapter 5

The AIMS home-video method: parental experiences and appraisal for use in neonatal follow-up clinics

BMC Pediatrics 2022; 22(1): 338

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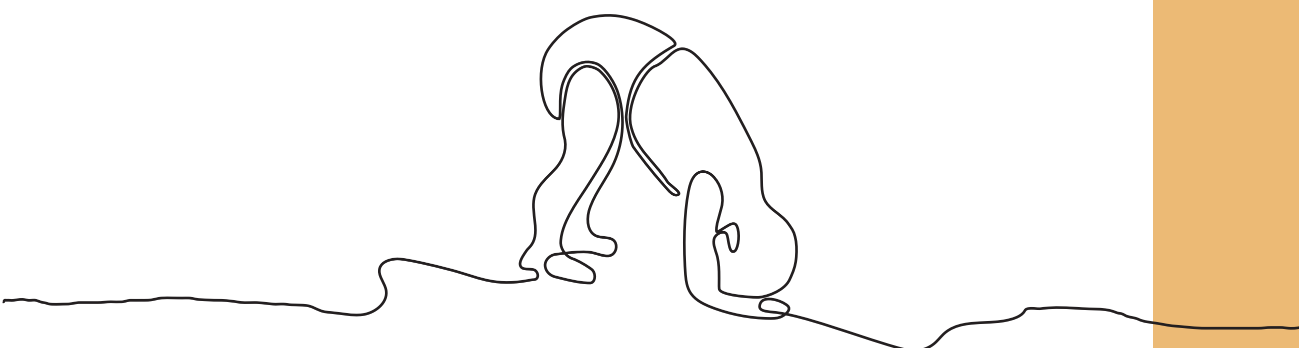

Abstract

Aim: Exploration of parental experiences with the Alberta Infant Motor Scale home-video method and their appraisal of its applicability for use in an outpatient neonatal follow-up clinic.

Method: A qualitative study among parents of healthy very premature infants (GA 26.2-31.5 weeks) participating in a longitudinal study of motor development between 3-18 months CA. Ten semi-structured interviews were conducted and transcribed verbatim. Inductive thematic analysis was performed following the process of the AIMS home-video method.

Results: Parents appraised the AIMS home-video method as manageable and fun to do. Transferring the video footage from their phone to their computer and uploading it to the web portal was sometimes time consuming. Parents gained a better awareness of their infant's motor development and found the feedback reassuring and a confirmation that their child is doing well. All parents thought that home-videos can be an addition to follow-up visits, but cannot replace (all) visits.

Conclusion: Parents appraised the AIMS home-video method positively and are of the opinion that home-videos can be of added value in monitoring infants at risk in neonatal follow-up additional to hospital visits. A secure and safe digital platform should be developed and implemented in neonatal follow-up, which should be explored in further research.

Key-words: AIMS, motor development, neonatal follow-up, very premature infant, eHealth

Introduction

Early screening and treatment of infants at risk is seen worldwide as an effective way of preventing health and social problems later in life [1–3]. Very premature born infants are infants at risk of developmental disorders, such as problems with gross and fine motor skills, problems with cognition, and social and/or behavioural problems [1,4-6]. Approximately 30% of these children experience problems with motor skills which often persist throughout childhood and sometimes into adulthood [6]. Early detection of developmental problems is therefore important.

In the Netherlands, between 2017 and 2019, approximately 7% of infants were born prematurely, of which 1.3% were born very or extremely prematurely (< 32 weeks gestational age (GA)) [7]. These infants are admitted to and looked after in hospitals with Neonatal Intensive Care Units (NICU). Because of increases in quality of care, the chances of survival of these infants has increased considerably over the past decades[4] After discharge, as advised by the European Standards of Care for Newborn Health[8] and according to the protocol from the Dutch Neonatal Follow-Up (LNF) Study Group for infants admitted at the NICU,[9,10] infants and their parents return to the hospital for regular check-ups at the follow-up clinic, where standardized tests are administered.
Using video footage to monitor infants might be a promising supplement to the check-up visits to the hospital. The use of eHealth technology may reduce costs, increase efficiency, provide easier access to health care and improve quality of treatment [11,12]. The need for remote care has become painfully relevant with the COVID-19 pandemic, resulting in many new solutions for providing and continuing care [13-15]. In recent years, many digital applications (apps) have been developed for health care purposes. These apps enable the monitoring of patients, provision of eHealth interventions and the collection of 'big data' [16,17].

5

Within the GODIVA-study (Gross mOtor Development of Infants using home-Video with the Alberta Infant Motor Scale), a method has been designed to assess an infant's motor development in which parents make a video recording of their infant at home, which is then assessed with an observational instrument, the Alberta Infant Motor Scale (AIMS). For longitudinal measurements of infants for research purposes, repeated filming has already been proved useful and feasible for parents of healthy term-born infants [18,19] Because it is often stressful for parents to have prematurely born infants at risk of developmental problems with subsequent need for medical care, the question arises as to whether parents of infants at risk find the AIMS home-video method useful for them as well [20]. In addition, home videos may contribute to monitoring infants at risk. The main purpose of this study was to gain an understanding of parental experiences of infants at risk with the AIMS home-video method. Subsequently, parents were asked how they appraised its applicability for use in an outpatient follow-up clinic.

Method

Study design

This study has a qualitative design involving semi-structured in-depth interviews with parents of very preterm (VPT) infants. Semi-structured interviewing offers participants sufficient opportunity to express their own views and helps to discover information not previously thought of [21].

Study setting

This study is part of a longitudinal study, the GODIVA-PIT study (to be reported on later). The GODIVA-PIT study (Gross mOtor Development of Infants using home-Video registration with the AIMS- following Premature Infants in Time) explores the motor trajectories of healthy premature infants (GA ≤32.0 weeks and/or with a birth weight <1500 grams) from 3.5 to 17.5 months corrected age (CA). Participants in the GODIVA-PIT study were recruited between May 2017 and December 2019 at the Wilhelmina Children's Hospital of the University Medical Centre Utrecht, Radboud University Medical Centre (Nijmegen), Isala Hospital (Zwolle) and by paediatric physical therapists of the TOP programme (Transmural developmental support for VPT infants and their parents) [22] throughout the Netherlands. Infants were recruited at regular neonatal or outpatient follow-up appointments, or during their first contacts with the TOP therapist.

Ethics

The GODIVA-PIT study was approved by the Medical Ethical Board of the University Medical Centre Utrecht (METC/UMCU), with reference number 17-186/C. Parents gave written informed consent prior to participation, in which they also gave consent to be contacted for another related study.

Sampling

Via convenience sampling, 20 families participating in the GODIVA-PIT study who had given permission in the Informed Consent to be contacted for other studies were approached, of which 10 agreed to participate **(Figure 1)**. The interviews were scheduled to commence after the parents had filmed their child at least once. When, after these 10 interviews, data appeared saturated, no further interviews were scheduled.

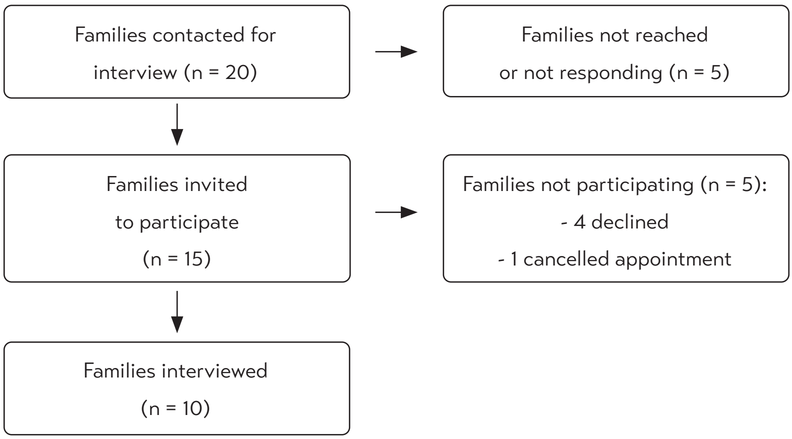

Figure 1: *Flowchart for participating families*

AIMS home-video method

In the GODIVA-PIT study, parents were asked to record their infant with the AIMS home-video method five to seven times with intervals of two to three months **(Figure 2)**. They received three instruction films and a booklet with three corresponding checklists. After parents had uploaded the videos via a secure web portal, the researcher and paediatric physical therapist (IS) assessed them with the AIMS and gave parents feedback on their infant's motor development by email. This email contained objective information on what was seen in the videos, a figure with norm references in which their infant's score was incorporated, and pictograms of the scores on the AIMS **(see Appendix I)**. Whenever abnormalities were seen in an infant's motor presentation, the attending physician and/or paediatric physiotherapist were contacted for consultation [23].

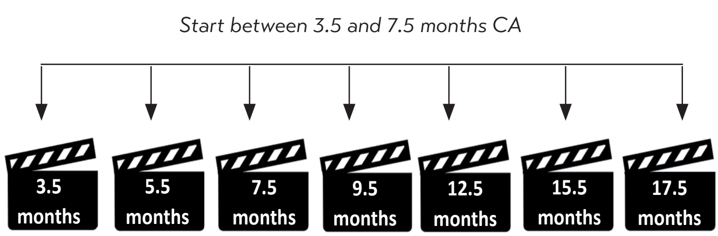

Figure 2: *Corrected ages of infants when recorded by their parents*

Data collection

Between January 2019 and February 2020 face-to-face in depth semi-structured interviews were conducted by a pair of interviewers, always consisting of the researcher (IS), who is also a lecturer on the master's programme Paediatric Physiotherapy, and a student of this programme (CW/AV/AS) who was under supervision of the researcher. The interviews took place in the family home. All interviews were video- and audiorecorded.

A guide with a topic list **(see Appendix II)** formed the basis for the semi-structured interviews. A pilot interview was conducted between researchers to test the interview guide. After each interview, deliberation took place with the two interviewers, and the guide evaluated and adjusted when necessary [24]. The guide provided key topics based on the comparable study of Boonzaaijer,[18] supplemented with topics regarding parents' views on using home videos for neonatal follow-up. Feedback on the topic list was provided by two experienced researchers (JN, MJ).

Data analysis

Audio recordings were transcribed verbatim according to a standard protocol. A thematic analysis approach [25] was used, guided by the research questions and the model of Boonzaaijer et al. [18] The phases of open, axial and selective coding were used for analysis to identify the most relevant themes [26]. The software program Atlas.ti was used for analysing and classifying the data [27].

Reliability and validity

To enhance reliability and validity of the data, all phases of the analysis were performed independently and compared afterwards. When no consensus was reached, a third researcher (JN) was consulted. During the first phase of the analysis, the researcher (IS) and two students (CW and KS) performed open and axial coding. In a second phase, all data were analysed by two researchers (IS and JO), including open, axial and selective coding. During analysis, a journal was kept with reflexive notes. Variation in the population was continuously monitored (i.e. fathers and/or mothers interviewed, infant GA, birthweight, number of times recorded). After nine interviews, data appeared saturated, which the last interview confirmed.
To enhance triangulation, three peer debriefing sessions were held with researchers and physiotherapists working in different fields (neurology, pain, psychosomatics and paediatrics), a paediatric health psychologist and a neonatologist. After these sessions, a final peer debriefing session took place to confirm the alterations in choices of quotations and names of the (sub)themes [26].

Results

We interviewed parents of 10 families: five interviews were conducted with the mother only, two with the father only, and three with both parents.
Mothers' median age was 34 years (range 28-40), fathers' median age was 35 years (range 30-45 years). Eight mothers and seven fathers were highly educated. Infants' median GA was 29 weeks (range 26.2-37.0), median birthweight was 1210 grams (range 960-2240). Parents filmed on average three times, with a range of one to seven times. One parent was a mother of twins, one parent had a post-migration background, and one infant was suspected of having cerebral palsy during the study. Parent and infant characteristics are shown in **Table 1**.

Table 1: Parental and infant characteristics

Interview	Sex	Parent(s) interviewed	Times recorded	Corrected Age infant at interview	Gravidity	Birth ranking	Health status	Parental country	Parental education (high[a]/ middle[b]/ low [c])[49]
1	boy	both	1	4 mo	singleton	1ste	healthy	Dutch	H/H
2	girl	mother (father came at the end)	2	9 mo	singleton	1ste	healthy	Dutch	H/H
3	girl	father	2	19 mo	singleton	3rd	healthy	Turkish	M
4	boy/ boy	mother	3	5 mo	twin	1st/2nd	healthy	Dutch	H
5	boy	mother	3	8 mo	singleton	3rd	healthy	Dutch	H
6	girl	father	2	9 mo	singleton	1st	healthy	Dutch	H
7	girl	mother	3[d]	13 mo	singleton	4th	healthy	Dutch	M
8	boy	mother	4	11 mo	singleton	1st	healthy	Dutch	H
9	boy	both	6[e]	22 mo	singleton	1st	healthy	Dutch	H/H
10	girl	both	7	20 mo	singleton	2nd	suspect of CP	Dutch	H/H

mo: month; CP: Cerebral Palsy a: high education = associate degree programs, higher education, Bachelor programs, Master degree programs, and doctoral degree programs b: medium education = upper secondary education, (basic) vocational training, and middle management and specialist education c: low education = primary school, prevocational secondary education, and lower secondary vocational training and assistant's training d: parents started participating in a study at the infants age of 5.5 months e: parents started participating in a study at the infants age of 7.5 months

5

The analysis will be presented in two parts, the first relating to the practical aspects of the AIMS home-video method together with the feelings and thoughts of parents using the method, and the second covering the parents' vision of the use of home videos in neonatal follow-up.

Figure 3 represents the overview of the practical aspects, and feelings and thoughts about the experiences with the AIMS home-video method. The practical aspects relate to the process of making the home video: the instructions, time planning, recording the video, uploading, and feedback. In **Table 2**, the extracted themes and subthemes are presented, accompanied by representative quotes

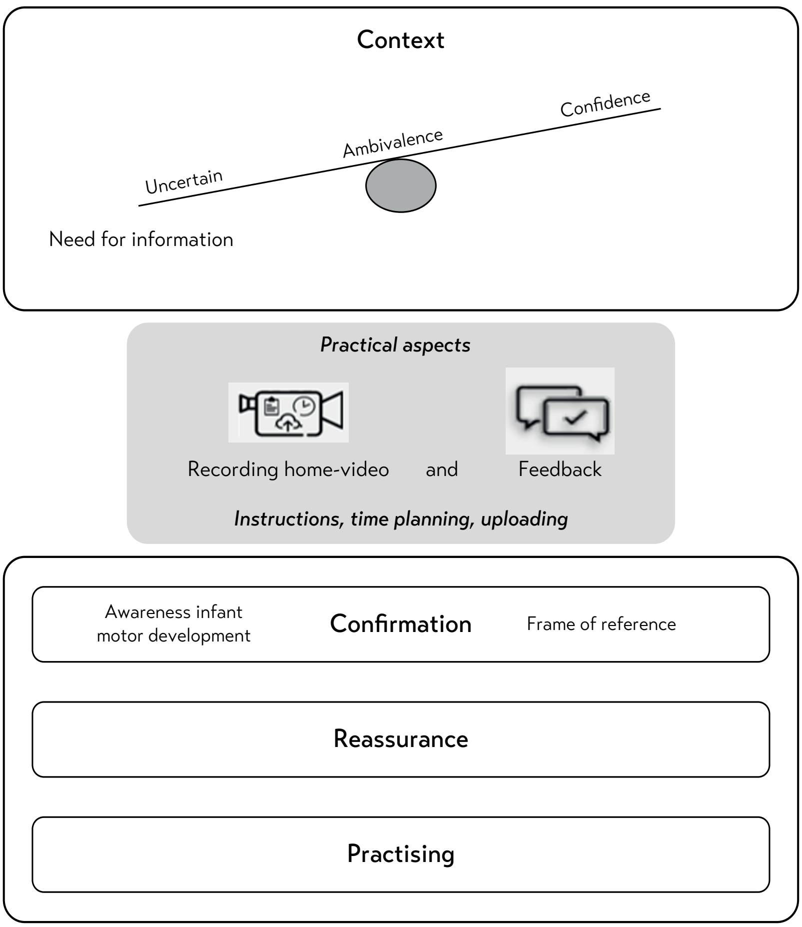

Figure 3: *Overview of the themes extracted from the interview data*

Part I: Parental experiences with the AIMS home-video method

Instructions

All parents considered the instructions on the checklists in the booklet ***clear***. Most parents watched the first instructional video on how they could film their infant, which was regarded as ***useful***. But it was not always clear that the three checklists entirely corresponded to the instruction videos. As a result of the instructions on the checklists, some parents actually started ***practising*** some of the items with their infant.

Time planning

This was the most challenging part of the home-video method. ***Recording on one day***, the necessity for ***two persons*** to record when the infant was young, undressing the infant and having the infant in ***the right state***, were perceived as most bothersome for recording. But parents also reported that recording ***became easier in time***, since:

1. parents knew what to expect of recording;
2. the infant slept less, so planning became easier;
3. the urge for two people to film reduced, due to improvement of the motor abilities of the infant.

Recording their infant in its ***own environment*** and choosing the right moment was appreciated and sometimes a prerequisite, or even the decisive factor, for participating in the study.

Recording home videos

Most parents experienced recording their infant as ***fun*** to do. Some parents said that prematurity made them more careful about handling their infant, when it was very young. Other parents mentioned that handling their infant for the video was ***similar to normal playing***. But if the infant was comfortable at the moment of recording, positioning the infant was not a nuisance.

During recording, parents occasionally discovered new motor skills in their infant, gaining a better ***awareness*** of their infant's motor development. In one interview with both parents, they said that because of the different recordings, one could actually see the development. Additionally, it made them more aware of what their infant already did than they would usually be during normal days.

Table 2: Quotations matching the themes and sub-themes regarding practical aspects, feelings and thoughts

Theme / Sub-theme	Quote
Instructions	
Clear	M: *'Yes, those (instructions) were pretty clear. Yes, with that (checklist), you really got [it].'* (int.8)
Useful	F: *'I learned … I saw how we had to record this film. So that was also useful.'* (int. 1)
Exercising	M: *'Sometimes we didn't know whether he could perhaps do certain things … or whether not. We thought, oh, perhaps that might be fun to offer [that activity] to him now. More like that. Because that's what we're going to do.'* (int. 5)
Time planning	
Recording on one day	M: *'You want [to do] it in just one day, of course, and that doesn't always work.'* (int. 3)
Two persons	F: *'Well, the limitation was that both of us had to film it.'* (int. 2) M: *'And filming that while standing, yes, I always need someone for that. And at a hectic pace, it does not always work smoothly.'* (int. 4)
Undressing	M: *'And you have to change his clothes a few times and I did not find that pleasant. He was actually tired by the time I had undressed him.'* (int. 5)
The right state	M: *'Yes, I usually plan it in my calendar. Then I think, oh, it is a day when we are both there, hey, on the weekend. But then he is just sleeping or then he has just been sick, [and] then the moment has passed.'* (int. 3)
Becomes more easy	M: *'It became shorter and shorter, I think, because he could actually do more [each time] and it was getting easier.'* (int. 10)
Own environment	M: *'That you don't again … because you are in the hospital quite a lot. I think that I would not have joined if I had to go somewhere [to take part]. I wanted to participate, because it could just be at home.'* (int. 4)
Recording home-video	
Fun	M: *'I actually found it very nice to do.'* (int. 2)
Similar to normal playing	M: *'You actually film what you already do with him every day.'* (int. 4)
Awareness MD	*F: 'It's nice that you see those different videos, that [motor] development. Then you are much more aware, I think. Otherwise you are not so aware of it day to day.' (int. 5)*

Uploading	
Time-consuming	*M: 'It really takes one or two hours (with transferring and uploading). So that is tough.' (int. 3)*
Feedback	
Frame of reference	*M: 'Especially with regard to how he is developing compared to other children of his age, corrected and not corrected [for his prematurity]. That's actually what I like most about it. And that you sort of look at how is he on the curve, is he going this way or that way. But above all, does it fall within the normal [range]?' (int. 3)*
Context	
Need for information	*M: 'And then I notice that I think, hmm, is that the way it should be? Or should he actually be able to [do that]? Or what is in it?' (int. 7)*
Uncertain	*M: 'But sometimes I find that difficult, because I don't... because I sometimes get insecure, because they are born too early.' (int. 7)*
Reassurance	*F: 'What else can go wrong, that was the hardest, I think. That matters a lot, in that it is nice to see again ... that we get confirmation that it's going well, orally, on paper and on screen.' (int. 9)*
Ambivalence	*M: 'I am very confident that I want my children, I want to stimulate them in their development, if that is necessary, but I also want them to actually do their own thing. Should I encourage them more because it's good for them, or should I let them do it themselves?' (int. 7)*
Confidence	*M: 'He [was] just born too early. ...so [there's] no reason why he shouldn't reach his milestones.' (int. 3)*
Confirmation	*M: 'It is a kind of confirmation of what you actually feel yourself.' (int. 6)*

M = Mother; F = Father

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Uploading

Most parents did not report any problems uploading the films to the web portal, although sometimes it was perceived as ***time-consuming***. However, some parents struggled with transferring the videos from their telephone to the computer. Suggestions for making uploading easier concerned mainly the user-friendliness of the web portal, e.g. by using an application on one's mobile phone.

Feedback

The feedback parents received was in general considered clear and good. The figure in the feedback **(Appendix I)** provided a ***frame of reference*** in which parents could see how their infant was developing, compared to peers. Interpretation of the graph with the norm references of term-born infants and premature infants was sometimes difficult, though the text below the figure and the explanation of the results in the email clarified this.

Generally, the feedback provided was a ***confirmation*** of what parents already thought about their child's development and, further, gave ***reassurance*** that their child was doing well. One father said that, while he knew what might go wrong in development due to the prematurity of his child, when he heard and saw that his child was doing well, he felt reassured. Besides, according to some parents, it was nice to have an extra pair of eyes monitoring their infant.

Context

Parents expressed the view that having a premature infant is stressful, with the realisation of having a different start with their infant than expected. The context of either being a first-time parent or already having more parenting experience also seems to matter.

Even at the time of admittance to the NICU, some parents had questions about their infant's development and felt the ***need for information***. Later, parents also had questions about what their child should be able to do at certain ages, and whether their child's actual repertoire was appropriate to their age. First-time parents seemed more ***uncertain***, reflected in feelings of doubt about their infant's development and hence a greater need for information about (motor) development. Recording their child made their infant's newly acquired motor abilities obvious, and feedback was found ***reassuring***. A few parents conveyed the impression of being inspired to practice with their child, according to the instructions. Although these parents created the impression of being more uncertain, some ***ambivalence*** emerged in that they also had

confidence in their child. The received feedback was often considered a ***confirmation*** of what they already thought about their child.

Parents who already had parenting experience seemed less uncertain and more confident about their infant’s development, reflected in having more faith in their infant's own pace in motor development. They reported less need for information and did not mention seeing new motor abilities, but expressed the need for comparison with their infant’s peers and for confirmation of what they already thought (i.e. that their child was doing well). Also, experienced parents did not mention practising with their child prompted by the instructions and/or recording.

<u>Deviant motor development</u>
The parents of the infant thought to have cerebral palsy reported similar themes despite differences in ***context*** where their child showed deviant motor development during the study.

These parents also became more ***aware*** and gained more knowledge about their infant’s motor development through ***recording*** their child and receiving ***feedback***. As a result of this feedback, they could see for themselves that their child deviated from the norm. This deviation reinforced the concern that their child was not developing as expected and was also a ***confirmation*** of what the doctor had said.

Because of the recording and feedback, the parents of the infant with the deviant motor development reported noticing more what their child could do, rather than what she or he could not do or should be able to do, according to standards. This may also be interpreted as ***reassuring***. Also, they were searching for a ***frame of reference*** for themselves, because the comparison with their older child was no longer valid.

Part II: Use of home videos for neonatal follow-up

Parents uniformly agreed that using home videos for monitoring infant motor development can certainly be an ***addition*** to follow-up visits but should ***not*** be a ***substitute*** for these. For instance, video recordings could be used in addition to regular check-ups when the doctor or parents themselves have questions about progress in other developmental domains, e.g. language or communication. In addition, parents consider using video footage as a way of providing information to other involved professionals, such as doctors in other hospitals or a speech therapist. Also, some parents saw the use of home videos as an opportunity to reduce the frequency of hospital visits, while still having their infant monitored.

On the other hand, parents emphasised the importance of doctors discussing with parents in person whether they wished to film their child: the importance and benefits of recording have to be clear at all times. Also, clear instructions, such as provided in the current study, should be given to parents on how and what to film.

Discussion

The present study describes the practical experiences, feelings and thoughts of parents of very preterm infants with the AIMS home-video method. In addition, parents gave their views on the suitability of home videos for use in outpatient follow-up clinics. Overall, parents found the AIMS home-video method to be manageable and fun to use, especially as infants get older; only transferring recordings from their phone to the computer and uploading them to the web portal was experienced as time consuming. Parents gained a better awareness of their infant's motor development and found the feedback to be reassuring, confirming that their child was doing well. All parents are of the opinion that home videos can be a useful addition, but not a replacement for, follow-up visits.

The GODIVA-PIT study was conducted in a similar Dutch (health care and cultural) context and used the same methodology as in the study of Boonzaaijer et al[18]. The main difference between the studies concerned the birth status of the children: while the current study included parents of preterm infants, that of Boonzaaijer et al. included parents of term-born infants. The majority of the (sub)themes in practical aspects and feelings and thoughts emerged in both studies, with only the content of the (sub)themes being different. In practical aspects, few differences arose, which may be explained by the improved digital capabilities of the mobile phones nowadays and the better functioning web portal (learning from previous errors). For instance, parents of the premature infants did not experience digital errors in uploading videos and low capacity for storage of footage in their mobile phones, unlike the parents of the term-born infants. Nevertheless, in both studies it often took a long time to upload the videos [18]. The major differences with the study of Boonzaaijer et al. are in the content of the (sub)themes of the feelings and thoughts, formed by the difference in the journey they have had in the birth of their premature infant. Parents of premature infants often experience a sudden disruption of the pregnancy, which makes them parents sooner than expected [28]. Next to this unexpected birth, the medical care is longer and more intensively accompanied with insecurities about their infant's wellbeing and future expectations than with healthy term-born infants [29]. When combined with becoming a parent for the first time, it seems natural to have feelings of uncertainty and to need information. That this uncertainty and need for information is less for parents of more than one child may be attributed to learning from experience, where parents use their experiences with their firstborns when faced with similar situations with subsequent children [30,31]. Experiences acquired with their firstborns increase their knowledge and effectiveness in meeting the needs and demands of later born children [32,19]. Parents appear to feel uncertain and vulnerable when they lack information of how to enhance their child's care [33]. In response to this uncertainty, it seems natural that parents of premature infants express their need for reassurance and confirmation that their child is doing well and that they are doing the right thing [29].

Interestingly, parental beliefs seem to play a role in expectations of development [34,35]. In our study, first-time parents felt that they should actively stimulate their child's motor development, while experienced parents were happy to trust their infant's own pace.

According to published research, parents in different cultures also differ in their beliefs about their infants' motor development and may therefore show differences in parental practices. For instance, first-time Israeli mothers of term-born infants attributed a bigger role to stimulation, whereas Dutch first-time parents attributed a bigger role to maturation and infants' own pace in development [35].

This study also gives insights into the appraisal of home videos for monitoring infants. Actively involving parents in neonatal follow-up perhaps contributes to Family Centred Care, which is supposed to enhance motor outcomes of the premature infants [36]. Recordings made parents aware of their infant's motor development, which may enhance empowerment which allows for increased confidence in parenting [37,38]. Giving feedback reassures parents and confirms how their child is doing, which may decrease stress levels in parents. These factors, empowerment and decreased stress, may contribute to (motor) development of the infant [20,36,37].

A relevant lesson learned from this study is that, when giving feedback, it is very important to tell parents what their child can do, as the parents of the infant with suspected cerebral palsy stated. It is important to concentrate on the strengths of a child, with positively phrased messages, and not just focus on weaknesses [29,30].

Strengths and limitations

Some limitations and strengths can be identified concerning the quality of the study. First, there was no member check to confirm whether the interpretation of the results as presented here was recognizable, which would have contributed to the internal validation of the data. Second, a convenience sample was used, which is more of a risk compared with a random sample. However, there appeared to be a sufficient reflection of the sample in parent (fathers and/or mothers interviewed), infant (GA, birthweight), and study characteristics (number of times recorded and therefore age of the infant during recording). On the other hand, there was only one infant with a deviant motor development. Parents gave different information, although almost all themes emerged in these interviews, though with a different content. A further point is that most parents were highly educated: research among Australian parents on the use of an application to assess infant general movements captured on a video made by parents showed that, while most parents used the Babymoves app successfully, parents of lower socio-demographic status used the app less [39]. Lastly, as in all research, the only parents participating were those interested in the study, which raises questions whether the AIMS home-video method is usable for monitoring all infants.

To increase validity and reliability of the interpretation of the data, the researchers endeavoured to be reflexive in the iterative process by making notes during the process and by independent coding. Arranging critical peer feedback and peer debriefing sessions where different perspectives on the data were involved enhanced triangulation.

Future research

Future research should aim at the implementation of a home-video method in neonatal follow-up. This would enable all professionals involved to get the same, realistic impression of an infant's abilities. The experiences of parents, as well as of the professionals involved, in using such a method should be explored. To enable this, a user-friendly application or platform to exchange video footage safely should be developed. During development, it is important to involve parents of different (post-)migration backgrounds, education levels, including parents of infants with deviant motor development.

Conclusion

Parents of preterm infants find the AIMS home-video method to be manageable, while receiving feedback reassures them and confirms that their child is doing well. Moreover, this method appears to be an intervention which enhances empowerment of parents in providing insight in their infant's motor development. It is suggested that home videos can be of added value in monitoring infants at risk in neonatal follow-up additional to hospital visits and to inform many of the health care professionals involved. A secure and safe digital platform should be developed and implemented in neonatal follow-up, which should be explored in further research.

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AUTHOR CONTRIBUTIONS

Imke Suir: Conceptualization, Methodology, Investigation, Resources, Data Curation, Formal Analysis, Writing – Original Draft, Project administration.
Janke Oosterhaven: Methodology, Formal Analysis, Writing - Review & Editing.
Marike Boonzaaijer: Conceptualization, Validation, Methodology, Writing - Review & Editing.
Jacqueline Nuysink: Conceptualization, Methodology, Validation, Formal Analysis, Writing - Review & Editing.
Marian Jongmans: Conceptualization, Methodology, Validation, Formal Analysis, Writing - Review & Editing, Supervision.

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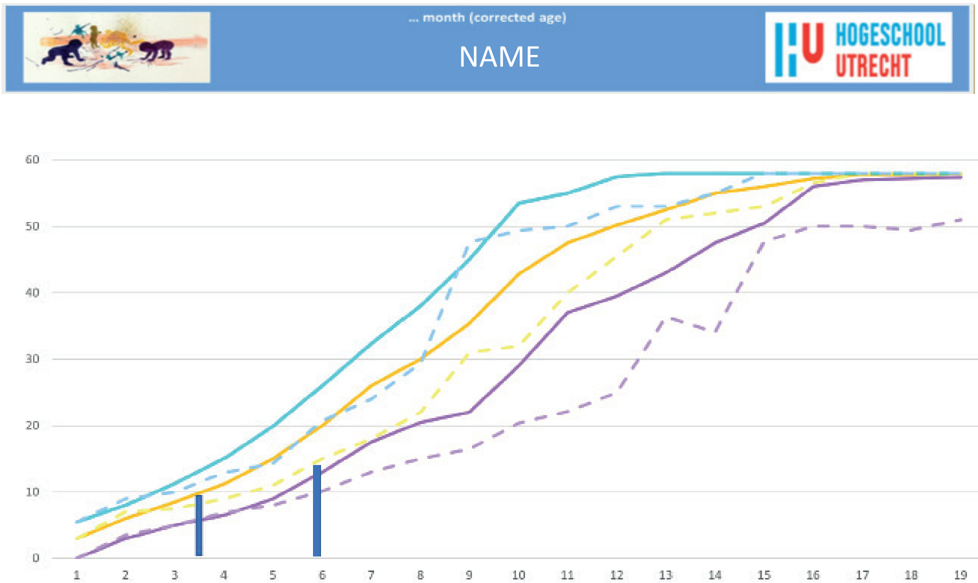
Appendix I: Example of the feedback to parents after an assessment.



Corrected age in months

Explanation 5th, 50th and 90th centile rank

In the graph above, your child's score is represented in relation to the 5th, 50th and 90th centile rank.

A centile score of 50 indicates that a child shows average motor development compared to peers.

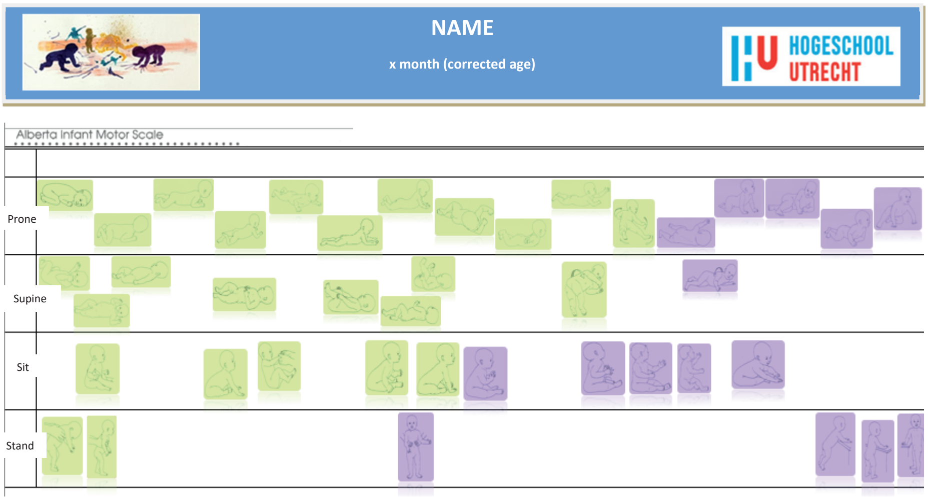
This score is a moment in time. It is an indication of where the child is in its motor development compared to peers at that time.

At the corrected age of x month, NAME falls between the 25th and 50th centile rank in respect to children born on time.

Compared to children born prematurely he/she falls between the 75th and 90th centile rank. This means that he/she scores according to the standards of motor development in comparison with peers who were also born prematurely.

Continues on next page

Appendix I: Example of the feedback to parents after an assessment. **(Continued)**



Explanation

Above you can see the score form of the instrument (AIMS: Alberta Infant Motor Scale) that charts motor development.

Based on the recordings of your child, we assessed the score and coloured the pictures which represent the items we score.

The green pictures display the items your child has shown in the video or has already mastered. The purple pictures are items your child did not show during the video.

The pictures are shown in chronological order of motor development. Because every child follows his/her own development, it is possible that your child skips an item.

To score a picture, specific requirements are set for the performance and posture. It is therefore possible that you think your child has shown a posture/movement which we have not been allowed to score, because the perfomance does not meet the requirements.

Appendix II: Interview guide

<table>
<tr><th colspan="3">Topic list</th></tr>
<tr><td colspan="3">Start</td></tr>
<tr><td colspan="3">• Thank you for your cooperation in this interview.
• Explanation of what is going to happen: “The purpose of the interview is to gain insight into the experiences of parents with the home video method. This makes it possible for us to improve and adjust the method. The interview is about your personal experiences with the GODIVA-PIT method. We would like to learn from your experiences. We are also interested in knowing how parents think about using videos made by parents, for doctor visits, like the neonatal follow-up visit, where you are going to with your child.
• Explaining some practical things: duration (about 45-60 minutes), recording equipment, anonymity.
• There are no right or wrong answers! What matters to us is your opinion. We want to learn from your experience.</td></tr>
<tr><td>Topic</td><td>Question</td><td>Extra questions</td></tr>
<tr><td>Start</td><td>• How did you like recording your child for this study?</td><td>facilitators and barriers</td></tr>
<tr><td>Time Planning</td><td>• Can you tell us the process of recording?
• Approximately how much time do you think you spent recording?</td><td>• organising/finding the right moment
• positive/negative aspects of recording at home
• time investment (every time, frequency, age of child)</td></tr>
<tr><td>Video footage (technically)</td><td>• What did you think of all the instructions?</td><td>• watching instructional videos
• read the instruction booklet
• checklists
• clear
• findability</td></tr>
<tr><td>Video footage (elicitation movement)</td><td>• What did you think of eliciting your child?
• How did you experience your participation in the study?</td><td>• performance child
• what you yourself have gained
• insight motor skills</td></tr>
</table>

Continues on next page

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Appendix II: Interview guide. **(Continued)**

Uploading	• What did you think about the uploading of the videos?	• manual/instructions for up-loading • uploading difficulties/problems • duration • expectation in advance • app?
Feedback	• What did you think of the feedback you got on the video footage?	• understanding feedback • influence feedback on handling
Follow up	• Do you think the video method is suitable for parents visiting the neonatal follow-up? • Why yes/no? • How do you envision using the video method for the neonatal follow-up?	• facilitators and barriers • added value
Prematurity	• What do you think of the video method for children born pre-maturely? • Can the method also be of added value for other children (with or without problems)?	• confronting • expectations own child
Expectations		
	• How stressful did you find re-cording your child for your child and yourself? • In the meantime, have you considered stopping the study? If so, why? • What motivated you to contin-ue the study?	

Continues on next page

Appendix II: Interview guide. **(Continued)**

Reflection		
	• What was your opinion about recording beforehand? • Was recording as expected? Why or why not? • What would you do differently next time? • What do you think could contribute that fewer parents drop out? • Do you have any questions about privacy? • Do you have anything to add to this interview?	

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Part III

Parents

Chapter 6

Parental beliefs about the motor development of Dutch infants born very preterm

Submitted

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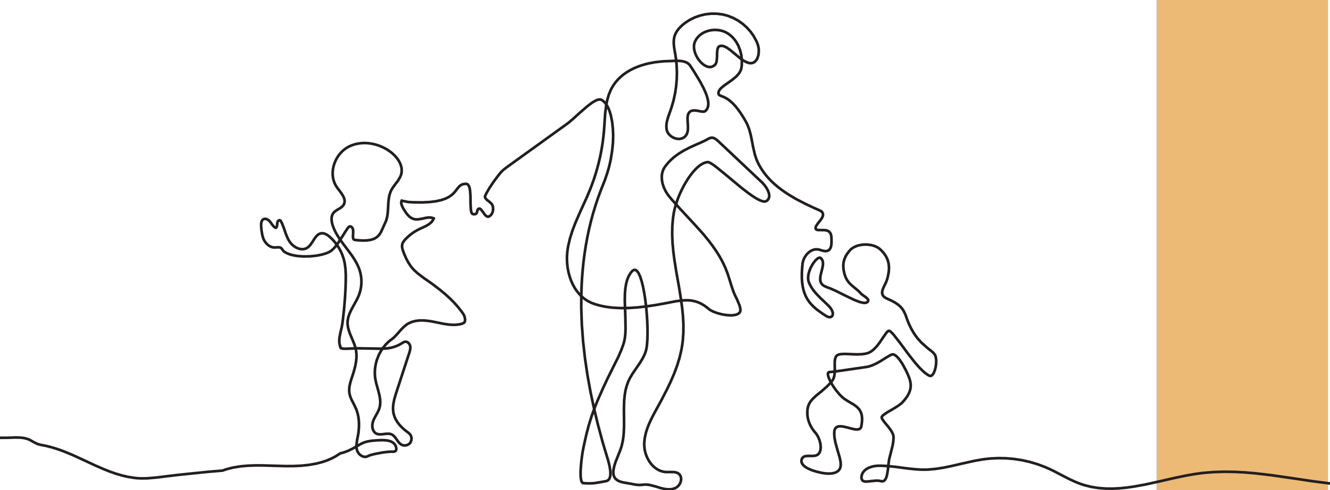

Abstract

Purpose: To explore the influence of preterm birth on parental beliefs about gross motor development and parents' supportive role in infants' motor development.

Methods: Prospective cohort study: Parents of very preterm (VPT) infants (gestation ≤ 32 weeks, birthweight < 1500 grams, without perinatal complications) and parents of healthy full-term (FT) infants completed the Parental Beliefs questionnaire.

Results: Questionnaires from 37 parents of VPT infants, aged 3.5—7.5 months (corrected), and 110 parents of FT infants, aged 3.5 months, were analyzed. VPT parents believed stimulating motor development to be more important than did FT parents ($F = 5.22$; $p = 0.024$; $p2 = 0.035$). Most VPT (82.4%) and FT (85.2%) parents acknowledged their role in supporting motor development. More VPT parents (41.2% vs 12.0%) believed they should follow their infant's natural developmental pace.

Conclusion: Knowledge of parental beliefs and parents' supporting role in motor development is relevant for tailoring pediatric physiotherapists' interventions with families.

Keywords: premature infant, parental beliefs, gross motor development, parental role

Introduction

Preterm birth is likely a stressful event for parents [1], with uncertainties about future developmental problems, including gross motor problems [2–5]. Development, in particular motor development, is more rapid in the first years of life than at any other age [6]. Infant motor development emerges in the interaction between factors within the infant and the environment [7]. The environment during the first two years is usually the infant's home, where the infant is completely dependent on its caregivers, mainly its parents, who therefore play an important role in its development [8]. Parental practices affect infant motor development by creating opportunities for the infant to develop and to explore the world [9,10].

The developmental niche is a theoretical framework describing the sociocultural construction of development [11]. Three subsystems, (1) physical and social setting, (2) parental beliefs about development and parenting, and (3) daily customs and practices of childrearing, interact with each other and with the developing child. Through this interaction, all subsystems may influence infant motor development. Research has already shown the influence of culture on gross motor development [12–14]. Still, little is known of parental beliefs about motor development.

Belief can be defined as the mental acceptance or conviction of the truth or actuality of an idea [13]. It is important to understand that beliefs can, but need not, be conscious: indeed, most beliefs are unconscious [15]. Beliefs serve as the basis for our understanding of the world around us and we therefore assume their truth. Beliefs have different origins and can vary in their impact on behavior [15]; one can act on certain beliefs and fail to act on others, depending on the degree of conviction and/or experience underpinning them. For example, beliefs are formed through past experiences and/or information from trusted sources [14]. Thus, it is plausible that parents confronted with preterm birth develop different beliefs to those of parents of full-term infants (FT). The stressful event of preterm birth and the risks of gross motor delays may alter parental beliefs and perceptions, influencing parenting practices [16]. Naturally, the information parents receive from involved healthcare professionals may also influence beliefs and/or behaviors.

Monitoring motor development is important for infants at risk. The pediatric physiotherapist (PPT) is often involved in monitoring and intervention during the first year of life of infants at risk. Family Centered Care (FCC) is considered best practice in early intervention [17,18], comprising active collaboration between the PPT and parents, respecting and honoring differences [19]. Consequently, for effective collaboration with parents, it is important to understand parental beliefs.

As well as understanding parental beliefs on motor development, it is important to gain knowledge of how parents consider their role in stimulating their infant's motor development. Parents' own perspective on their role may affect parenting behaviors [20], but little is known about this.

The aim of this study is to explore whether and, if so, how preterm birth influences parental beliefs about motor development and how parents consider their own role in the motor development of their infant.

This led to the following research questions:
1. What are the similarities and/or differences in parental beliefs about motor development between parents of Dutch VPT and FT infants?
2. Do parents of VPT and FT infants differ in their beliefs about their own supportive role in their infant's motor development?

Methods

Study design

This study was part of a large prospective cohort study, GODIVA (Gross mOtor Development of Infants using home-Video registration with the Alberta infant motor scale (AIMS)). The overall aim of the GODIVA study was to better understand infant gross motor development from birth until independent walking and the factors related to the shape and speed of gross motor developmental curves. Two longitudinal sub-studies were initiated, with the first following FT infants from 3.5 to 15.5 months of age (GODIVA-KIT study) and a subsequent one following VPT infants from 3.5 to 17 months corrected age (GODIVA-PIT study). For the current study, data from both sub-studies are used. Part of the GODIVA-KIT data has previously been used to answer a different research question concerning the change over time in parental beliefs about gross motor development of FT infants [21].

Participants

For the GODIVA-KIT study, parents of FT infants (FT parents) were recruited between May 2016 and April 2018 through open registration. Infants were recruited by dis-

tributing flyers at birth centers, day-care centers, well-baby clinics, and maternity care offices in the larger cities of the Netherlands. Infants were excluded from the study if they were born before 37 weeks gestational age (GA) or diagnosed with pathology.
For the (current) GODIVA-PIT study, parents of VPT infants (VPT parents) were recruited between May 2017 and December 2019 in the Wilhelmina Children's Hospital (University Medical Centre Utrecht), Radboud University Medical Centre (Nijmegen), Isala Hospital (Zwolle) and by TOP (Transmural development support for VPT infants and their parents) PPTs throughout the Netherlands [22]. Infants were recruited at the regular neonatal follow-up, or during their first contact with the TOP PPT. Most parents of infants in the Netherlands born before 32 weeks gestation and/or weighing less than 1500 grams are advised to participate in the TOP program. Eligible infants were born before or at 32.0 weeks GA or with a birthweight (BW) of <1500 grams and younger than 7.5 months (corrected for preterm birth) at the start of the study. Their parents had to understand Dutch language sufficiently. Infants were considered ineligible if diagnosed with a known syndrome, a neuromuscular disorder, severe neuroimaging abnormalities (e.g., cystic periventricular leukomalacia, IVH grade III or IV), meningitis, bronchopulmonary dysplasia (defined as oxygen supplementation >36 weeks postmenstrual age), congenital anomalies, necrotizing enterocolitis requiring surgical procedures, prolonged tube feeding (defined as beyond hospital discharge), and severe visual or hearing disorder.

Procedures and measures

When infants met the inclusion criteria, parents were asked to participate and received information accompanied by a request for informed consent. After approximately a week, parents were contacted to answer any questions and asked to return signed consent forms if they agreed to participate. Booklets with information, checklists, and instructions were sent to them. The GODIVA-KIT and GODIVA-PIT studies had similar protocols in which parents were asked to record their infant six times with the Alberta Infant Motor Scale (AIMS) home-video method [23] if their infant was full-term and seven times if preterm. In addition, before parents in both studies started filming, they received a demographic questionnaire, and the Parental Beliefs on Motor Development (PB-MD) questionnaire [24] by email, being asked to fill this out before the first time recording their infant. FT parents received the questionnaire when their infant was 3 months old. VPT parents received the questionnaire when their infant was 3, 5 or 7 months corrected age (CA).

Measurement

The Parental Beliefs on Motor Development (PB-MD) questionnaire comprises four sections. The first section includes seven statements and the second four case descriptions, followed by statements representing possible interpretations and approaches. In these two sections, parents rate their agreement with the statements on a 6-point scale from 1 (disagree) to 6 (strongly agree). Factor analysis of the first two sections reveals a single item indicator and five scales measuring such beliefs as: 1) stimulation of motor development is important; 2) motor development occurs naturally; 3) seeking advice on motor development is important; 4) order of motor development is important; and 5) children should follow their own pace in motor development24. Scale scores are calculated from the means of the corresponding scale items (recoded where needed). The third section contains two open-ended questions on ideas about parenting, specifically how parents consider their role in their infant's gross motor development and whether parents think they should do something to support this. The fourth section, on sources of information about motor development, was not part of our study. The reliability and validity of the PB-MD are good [25].

Ethics

Both the GODIVA-KIT and GODIVA-PIT studies were approved by the Medical Ethical Board of the University Medical Centre Utrecht (METC/UMCU) with protocol nos. 16/366C and 17-186/C respectively. Parents gave written informed consent prior to participation. Video data were stored on a secure server at Utrecht University of Applied Sciences.

Data analysis

Sample characteristics were calculated with descriptive measures. Chi-squared and Fisher's exact tests were performed to compare most infant and parent characteristics between the VPT and FT infants. Cramér's V or Cohen's d were calculated for the effect size, with $d < 0.2$ being small, 0.2-0.7 medium, and > 0.8 large effect sizes [26–28]. Only for BW and GA were independent Student's t-tests calculated for effect sizes. Scale scores were calculated by averaging the sum of the scale items.

The single-item indicator and scale scores were tested for normality, considering normality to obtain when skewness and kurtosis were between -2 and 2. The single-item indicator and the scale scores (Sections 1 and 2) were normally distributed and therefore a MANOVA analysis was conducted on the differences between VPT and FT parents. Partial eta squared (p2) values were calculated for the effect of the differences. For all tests, a p-value of <0.05 was considered significant. For p2, effect sizes >0.01 are considered small, >0.06 medium and >0.14 large [26,27,29].

The two open-ended questions (Section 3) were coded using a previously developed coding scheme **(see Appendix 1)** [24]. After training, the researchers of the GODIVA-PIT (IS) and -KIT (MB) study and five Master's PPT students (SW, AI for GODIVA-PIT; DW, MW, JM for GODVIA-KIT) independently coded the two open questions in pairs, together with one of the researchers. Identified codes were rated as dichotomous outcomes (yes = 1; no = 0). The percentage of parents who mentioned a code was calculated. Differences between percentages were calculated with Chi-squared or Fisher's exact tests, with Cramér's V analysis for effect sizes.

Results

Demographics of the VPT and FT samples

Data from the demographics questionnaire of 37 VPT parents and 110 FT parents were analyzed and compared (**see Table 1**). Infant characteristics were, as expected, only significantly different for their GA and BW. Parental characteristics differed only in paternal education, where fathers of FT infants had a higher educational level. Significantly more VPT infants ($p = <0.001$; Cramer's V = 0.919) had one or two parents with a non-Dutch mother tongue. Also, more VPT infants had received pediatric physical therapy than FT infants.

Comparison of parental beliefs between VPT and FT parents

To answer the first research question, a multivariate test was performed which only showed a significant difference between parents on the Stimulation scale, albeit with a small effect size ($F = 5.221$; $p = 0.024$; ∏p2 = 0.035). This implies that VPT parents agreed more with stimulation of motor development than FT parents **(see Figure 1)**. Despite the significant difference between VPT and FT parents, on average both groups tended to disagree with belief in stimulating motor development (VPT mean score = 2.8 (±0.8); FT mean score = 2.5 (±0.7)).

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Table 1: Demographics and comparison of VPT and FT sample characteristics.

	VPT infants (n = 37)	FT infants (n = 110)	Sig. (p)	Effect size (∏p²)
Infant characteristics				
Sex (girls)	16 (48.6%)	66 (60%)	0.155[a]	0.100[c]
GA (weeks)			**< 0.001[b]**	**6.876[d]**
Mean (±SD)	29.5 (±2.1)	39.9 (±1.12)		
BW (grams)			**< 0.001[b]**	**5.532[d]**
Mean (±SD)	1198 (±341)	3556 (±451)		
Birth order			0.211[b]	0.145[c]
1st	25 (67.6%)	56 (50.9%)		
2nd	9 (24.3%)	40 (36.4%)		
3rd or more	3 (8.1%)	14 (12.7%)		
Pediatric Physical therapy / TOP			**< 0.001[a]**	**0.758[c]**
Yes	34 (91.9%)	12 (10.9%)		
No	3 (8.1%)	98 (89.1%)		
Parental characteristics				
Maternal age (years)			0.780[b]	0.109[c]
≤24	0 (0%)	2 (1,8%)		
25-29	7 (18.9%)	17 (15.5%)		
30-34	21 (56.8%)	55 (50.0%)		
35-39	7 (18.9%)	28 (25.5%)		
≥40	2 (5.4%)	8 (7.3%)		
Paternal age (years)			0.579[b]	0.161[c]
≤24	0 (0%)	1 (0.9%)		
25-29	3 (8.1%)	15 (13.6%)		
30-34	15 (40.5%)	32 (29.1%)		
35-39	16 (43.2%)	44 (40.0%)		
≥40	3 (8.1%)	16 (14.5%)		
unknown	0 (0%)	2 (1.8%)		
Maternal education			0.282[b]	0.161[c]
No education	0 (0%)	0 (0%)		
Primary	1 (2.7%)	0 (0%)		
Secondary lower	0 (0%)	2 (1.8%)		
Secondary higher	6 (16.2%)	15 (13.6%)		
Tertiary	30 (81.1%)	93 (84.5%)		

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Table 1: Demographics and comparison of VPT and FT sample characteristics. **(Continued)**

	VPT infants (n = 37)	**FT infants** (n = 110)	**Sig. (p)**	**Effect size (∏p^2)**
Paternal education			**0.008[b]**	**0.305[c]**
No education	0 (0%)	2 (1.8%)		
Primary	0 (0%)	1 (0.9%)		
Secondary lower	5 (13.5%)	1 (0.9%)		
Secondary higher	8 (21.6%)	16 (14.5%)		
Tertiary	24 (64.9%)	90 (81.8%)		
Parental language	30 (81.1%)	105 (95.5%)	**< 0.001[a]**	**0.919[c]**
Dutch	7 (18.9%)	5 (4.5%)		
Other than Dutch				
Parents filling out questionnaire				
Age (CA) of infant		110 (100%)	**< 0.001[b]**	**0.386[C]**
3.5 months	30 (81.1%)			
5.5 months	3 (8.1%)			
7.5 months	4 (10.8%)			

a = Fisher's exact test; b = Chi-squared test; c = Cramér's V; d = Cohen's d

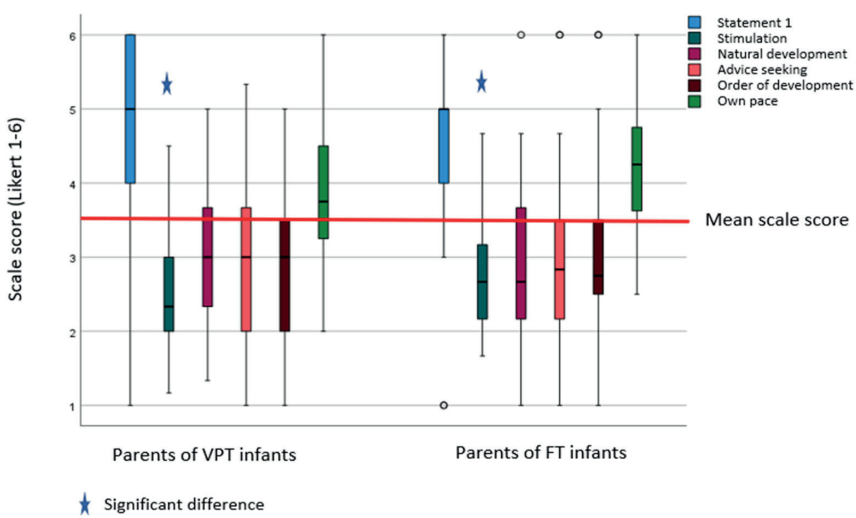

Figure 1: *Box plot with the comparison of the scores on the first statement and the scales of the PB-MD between VPT and FT parents.*

The scale Own pace was marginally significant, showing a small effect size (F = 0.012; p = 0.080; p2 = 0.021). The first statement (which is the single item indicator) and the scales Natural development, Advice, and Order of milestone attainment did not reveal significant differences between the parents.

Though not significant, all parents expressed a strong belief that motor development is most important in the first year of life, with a mean scale score of 4.6 (±1.1) for VPT and 4.8 (±1.1) for FT parents

Table 2: MANOVA of the comparison between VPT and FT parents on the scales of the PB-MD.

	Mean (SD)		B	F	SE (total)	Sig. (*p*)	Partial eta Squared (∏p2)
Dimension	**VPT**	**FT**					
Statement 1	4.7 (1.2)	4.9 (1.1)	0.253	1.403	0.213	0.238	0.010
Stimulation	2.8(0.8)	2.5(0.7)	-0.302	5.221	0.132	**0.024***	0.035
Natural development	2.9 (1.1)	3.0(0.9)	0.102	0.293	0.188	0.589	0.002
Advice	3.0 (1.1)	2.8 (1.0)	-0.139	0.473	0.202	0.439	0.003
Order	2.4 (1.3)	2.7 (1.3)	-0.109	0.350	0.185	0.555	0.002
Own pace	4.1 (1.0)	3.9(0.8)	-0.285	3.115	0.162	0.080	0.021

a Partial et squared

**Significant difference (p < 0.05)*

Parental role regarding motor development

To answer the second research question about parents' role, the two open questions were analyzed. **Table 3** shows that most parents answered 'yes' to the question of whether parents have a supporting role in their infants' motor development (VPT parents 82.4% vs 85.2% FT parents). However, some parents felt that, thoughthey had a role, it was not their goal to accelerate motor development **(see Quote 1 in Table 4).**

Table 3: *Percentages of parents of VPT and FT infants describing their parental role and providing activities concerning their infants' motor development.*

		VPT n = 34	VPT %	FT n = 108	FT %	Sig. (p)	Effect size
Parental role:	In general: Yes, support	28	82.4%	92	85.2%	0.437a	0.066
	No need to support / No need to compare to other children / children follow their own (natural) pace of development / the baby will develop the skills regardless of support	9	26.5%	29	21.2 %	0.896a	0.011
	Signal /encourage when necessary / consult experts / observe problems	6	17.6%	25	23.1%	0.453a	0.064
	Follow child according to age norms / or according to child's abilities / follow the child's (natural) developmental pace	14	41.2%	13	12.0%	**< 0.001**[a]	0.313
	Support, but not over-support/ don't push /overstimulate	11	32.4%	22	20.4%	0.175[a]	0.115
	Foster motor development, by right environment/ toys/ equipment/ reward / light stimulation / elicit motor development	5	14.7%	25	23.1%	0.262[a]	0.095
	Encourage / mild stimulation	4	11.8%	6	5.6%	0.259[b]	0.101
	Active stimulation	0	0%	1	0.9%	1.000[b]	0.048

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Table 3: Percentages of parents of VPT and FT infants describing their parental role and providing activities concerning their infants' motor development **(Continued)**

		VPT n = 34	VPT %	FT n = 108	FT %	Sig. (p)	Effect size
Activities:	Manipulate movement / active stimulation	0	0%	18	16.7%	**0.007[b]**	**0.219**
	Putting in prone	10	29.4%	25	23.1%	0.513[a]	0.055
	Putting in sitting	0	0%	2	1.9%	1.000[b]	0.069
	Putting in standing	0	0%	2	1.9%	1.000[b]	0.069
	Stretch	0	0%	0	0%		
	Help rolling	1	2.9%	4	3.7%	1.000[b]	0.020
	Massaging	0	0%	1	0.9%	1.000[b]	0.048
	Doing exercises	2	5.9%	1	0.9%	0.148[b]	0.146
	Stimulate senses	1	2.9%	0	0%	0.245[b]	0.150
	Other manipulating movements	4	11.8%	9	8.3%	0.520	0.047
	Adapting activities to age norms / or adapting activities to the child's abilities	7	61.7%	47	43.5%	0.085[a]	0.146
	Foster/facilitate through environment/toys / light stimulation	21	61.7%	47	43.5%	0.085[a]	0.146
	Other activities, general or not motor	12	35.3%	42	38.9%	0.625[a]	0.042
Parents' outdoor activities in manipulating movement:	Baby swimming	2	5.8%	9	8.3%	1.000[b]	0.043
	Baby yoga	0	0%	2	1.9%	1.000[b]	0.069
	Other movement classes	0	0%	7	6.5%	0.194[b]	0.131

a = Pearson's chi-squared test

b = Fisher's exact test

Table 4: Quotes from the two open questions.

Quote 1	"Yes. Not to make it go more rapidly but not to slow down growth. She (their daughter) loves practicing and getting undivided attention. Facilitator and guide." (parent of a VPT infant)
Quote 2	"Yes, parents can help the child by playfully stimulating the motor development that the child is currently capable of." (parent of a VPT infant)
Quote 3	"Yes, but don't force it and practice something for too long" (parent of a FT infant)
Quote 4	..."In the first weeks, parents should also be alert to the child's [sleeping or lying] position, in case of a possible preferred position. The child will not 'solve' this on its own"....(parent of a FT infant)
Quote 5	"Lots of practice (laying on the tummy), challenge and stimulation can speed up the process, but you can never have this one hundred percent guaranteed." (parent of a FT infant)
Quote 6	"...e.g. offering toys, providing space and opportunity to engage in motor activities (laying on tummy, putting in the playpen/on a play mat)." (parent of a VPT infant)

Few differences were found between VPT and FT parents considering parents' role in stimulating motor development. One was that more parents of VPT infants (35.3% VPT parents vs 9.3% FT parents; $p < 0.001$; Cramér's V = 0.363) stated that their role was to follow the child's (natural) developmental pace **(see Quote 2 in Table 4)**.
More than 30% of VPT parents and 20% of the FT parents said that parents should support infant motor development, but not over-support or push the infant **(see Quote 3 in Table 4)**. Some parents (17.6% VPT parents vs 23.1% FT parents) said they had a signaling role **(see Quote 4 in Table 4)**. Remarkably, only one of the FT parents described the role of parents as actively stimulating their infant **(see Quote 5 in Table 4)**.

In answering the second open question of whether parents should do something with the baby and/or environment to support infant motor development, most VPT parents (61.7%) gave answers about fostering or facilitating by creating the right environment, and providing right toys, space and/or equipment. Of the FT parents, 43.5% also reported this ($p = 0.085$; Cramér's V = 0.146) **(see Quote 6 in Table 4)**.

Of the FT parents, 16.7% reported actively stimulating their infant, while none of the VPT parents gave that answer ($p = 0.007$; Cramér's V = 0.219). Both groups of parents described different activities they provided for their infant. The most commonly described activity for stimulating their infants was putting in prone position (VPT parents 29.4% vs FT parents 23.1%).
There were no significant differences in the activities parents provided for their infants, although only twice (5.8%) did VPT parents report going to baby swimming; in total, FT parents reported providing infant activities 18 times (16.7%) (baby swimming, yoga and/or other movement classes).

Discussion

Because research on parental beliefs about motor development and how this eventually affects the actual gross motor development of infants is still scarce, the aim of our research was to gain a better understanding of parental beliefs and supporting role in the gross motor development of Dutch VPT and FT infants. This study demonstrated that there were few differences between Dutch parents of VPT and FT infants in their beliefs about motor development. While VPT parents, as with FT parents, agreed that motor development is one of the most important things during the first year of life, they do not hold clear beliefs in favor of actively promoting motor development, though they believed more than FT parents that stimulating motor development was important.

Most parents, both VPT and FT, believed that stimulating motor development was the role of a parent. VPT parents believed more that their role was to follow the infant's natural pace of development, rather than actively stimulating motor development. VPT parents more often reported stimulating their infants by creating the right environment and/or using toys or equipment.

Though significant, the difference between VPT and FT parents on stimulating motor development, where VPT parents believe more in stimulating motor development, is small. This difference might be related to the fact that VPT infants almost all receive TOP therapy, performed by a PPT. On the other hand, parents of FT infants visit baby clinics where motor development is also screened, but the accent of that visit is perhaps different. Despite this difference, for both sets of parents the average scale score is less than 3.5 which means that parents do not strongly believe that they should stimulate their infants, and even tend toward a belief that they should not. That Dutch parents tend to believe less in stimulating motor development of their infant is in line with research on the differences between the beliefs of Israeli and Dutch parents. Research on parenting beliefs in Western cultures has already shown cultural differences[30–32]. Cross-cultural research between Israeli parents and Dutch parents with the PB-MD questionnaire reveals that Dutch parents overall tend to believe less in stimulating motor development than do Israeli parents [30].
VPT parents, as well as FT parents, believe that motor development in the first year is very important, with a mean score on this item of more than 4.5. On the other hand, VPT parents in particular do not tend to actively stimulate motor development, rather believing that their infants will develop these skills regardless of support.

There seems to be some contradiction between parents' beliefs about motor development and their practices. One might expect that if motor development were seen as important in the first year of life, parents would act accordingly, i.e., stimulating their infants' motor development. However, beliefs do not seem to naturally align with practices. Possibly, the circumstances of preterm birth and all that comes with it may change the way they interact with their infant, but may not change their core beliefs, with the difference between beliefs and practices becoming bigger in these more extraordinary circumstances.

Our study did not find large differences between VPT and FT parents, which may reflect the idea that within the same country the same cultural model exists. This may lead to the assumption of homogeneity in parenting with Dutch parents. On the other hand, though small, real differences are found between VPT and FT parents. However, this may merely imply that, even though the PB-MD is a valid and reliable questionnaire to measure differences between cultures, it is less suitable to do so within one culture.

Another finding was that, compared to FT parents, VPT parents more often see their role as following the child in its own developmental pace. One possible explanatory factor for this result is that almost all VPT infants receive TOP pediatric physiotherapy. Because the TOP program is a preventive responsive parenting program for VPT infants and their parents[22], the latter are perhaps more alert to their infants' abilities and do feel the need to stimulate their infant actively. This is in line with the finding that, although only marginally significant ($p = 0.085$), VPT parents say their role is to promote motor development more, by creating the right environment, toys, equipment, etc.

It was apparent that VPT parents were less in favor of active stimulation. Parents often think of premature infants as more vulnerable[33]. When an infant is considered more vulnerable, it may be that VPT parents think stimulating their infant is less important, for fear that one might ask too much of the infant. Our results show that parents of preterm infants do not go out much for activities like baby swimming, baby yoga, etc., possibly because they consider their child more vulnerable. At the Wilhelmina Children's Hospital, parents are advised to be cautious about taking their VPT infant to the daycare center because of the higher risk of respiratory infections in the first year. Perhaps parents follow this advice in a broader way.

Limitations of the study

The first limitation is the small sample size of VPT infants, which makes generalizing results and conclusions more difficult.

Secondly, the FT infants were approximately 3 months old, but the VPT infants were 3.5 to 7.5 months CA and therefore at least 5 months calendar age. Besides, almost all VPT infants (92%) had received pediatric physiotherapy, compared with only 11% of FT infants. Since beliefs are formed based on past experiences and/or information from trusted sources14, it is possible that changes in beliefs, based on the experience of preterm birth and a minimum of five months of caring for VPT infants, and accompanying information from health care providers, may partly explain the difference between VPT and FT parents (although this was not part of this study). This may make comparisons between the two groups difficult. On the other hand, the parent groups are necessarily unequal in their experiences with their infants, and little is known about changes in beliefs and the constructs behind them.

Clinical implications

For PPTs, it is useful to understand parental beliefs about gross motor development in infants that need intervention. If parents feel motor development to be important in the first year of life but that they do not have to stimulate their infant (which will develop at its own pace), this is valuable information for professionals. If parents have concerns about their infants' motor development, PPTs can give more information on the relevance of stimulating gross motor development. In general, it is valuable to know what parents consider their role to be: for instance, if they believe that their role is to create a stimulating environment, the PPT can respond to this appropriately.

Future research

For research, the PB-MD is a valid and reliable questionnaire to gain more insight into parental beliefs cross-culturally[24]. Intra-culturally, the questionnaire may not be sensitive enough to compare different groups of parents. Moreover, the use of the questionnaire in clinical practice has not yet been tested. As the questionnaire was designed for research, it is not obvious that it is applicable in clinical practice, or even that a questionnaire is the best approach for identifying parental beliefs. Therefore, intra-cultural research on parental beliefs and on tools for identifying parental beliefs in clinical practice is required.

Also, more research into changes of parental beliefs and their influence on parental practices would contribute to a better understanding of what PPTs may be able to contribute during interventions with VPT infants.

Conclusion

Few significant differences were found in parental beliefs between Dutch parents of VPT and FT infants, perhaps explained by their sharing the same cultural context. Identification of differences in beliefs within the same culture may require a different approach to that of the PB-MD questionnaire. Knowledge of parental beliefs about gross motor development and how parents consider their own supporting role in this, though relevant to PPTs, is scant. Such knowledge would provide possibilities for PPTs to relate to parents and their beliefs regarding gross motor development, helping them to adapt to the parents' needs and practices.

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AUTHOR CONTRIBUTIONS

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

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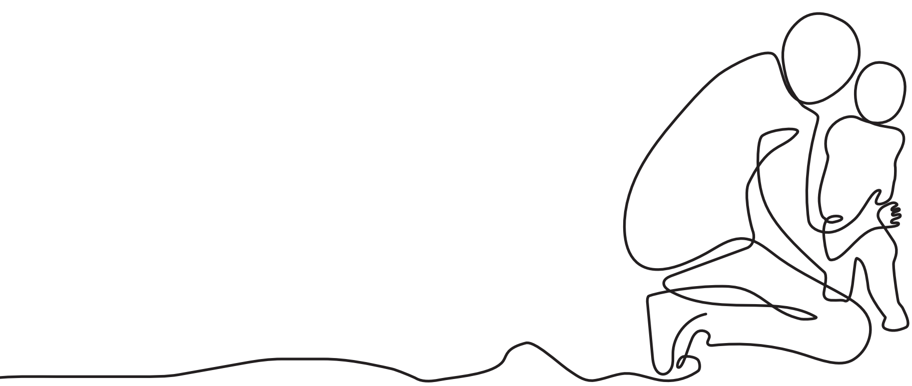
Chapter 7

Summary of findings and General Discussion

Imke Suir



Scope of the thesis

Parents are visiting the PPT for the last time, with their 18-month-old (CA) infant. They are very happy to visit the PPT because their son -born with a GA of 29 weeks, a BW of 1356 grams, and a five-minute Apgar score of 6- just started walking independently and they want to show this to their PPT. In the past 16 months, they have seen the PPT on a regular basis. The PPT always started with an observation of their infant, looking at what he had already achieved in his motor development. Every few months, she measured his gross motor development with the AIMS. The first time the PPT assessed their infant with the AIMS, parents were quite shocked by his low score. The PPT reassured them right away and indicated that one assessment is a snapshot and provides only an impression of their infants' gross motor development for the short term. Throughout the follow-up period, multiple assessments were performed, and he scored below the lowest norm (5th percentile) a few times, but not consistently. Over time he appeared not to be the fastest in his gross motor development but nevertheless made steady progress. During the COVID-19 pandemic, national restrictions prevented him from seeing the PPT. Parents were asked to use the AIMS home-video method instead so that the PPT could continue to monitor their infant's progress during this period. The PPT always emailed feedback to the parents explaining how their infant was doing. Parents became more aware of what their infant was capable of and were reassured that he was doing well. Being able to compare the AIMS scores with other premature infants, their initial concern about his motor development disappeared; he appeared an average developer. Combined with all other aspects of his development, they saw that he was doing well. Besides the fact that each infant develops at its own pace, and one should just provide an environment challenging an infant's capabilities, the PPT explained that their infant needed more stimulation and support in its motor development and gave advice on how parents could do this. As a result, he now walks independently, though sometimes stumbling and falling. Yet he can explore the world around him from a new, upright, viewpoint.

The case presented at the start of the general introduction of this thesis and above illustrates daily routines of the PPT to whom parents come with many questions. The case presented above is of course one of the many possible outcomes.
This thesis aimed to provide PPTs (and/or other clinicians) and parents knowledge and tools to support clinical decision-making and shaping early interventions.

Summary of the main findings

The first aim was to examine whether the currently used Canadian AIMS norms are appropriate for infants in the Netherlands **(Chapter 2)**. With the AIMS home-video method, 499 parents of typically developing infants recorded their infant once. The age of the infants ranged from 2 weeks to 19 months after birth. The distribution of the number of infants in the monthly age categories was comparable to the Canadian reference study, with fewer infants at the start and end of the age categories. Using the scaling method, we calculated item locations of the AIMS items, and compared the Dutch sample to the Canadian sample. Of the 58 items, 45 items met the criterion for analysis. Results revealed that in 42 out of these 45 items Dutch infants passed these at an older age. Also, monthly age groups of Dutch infants had lower mean AIMS raw scores compared to Canadian infants. Still, the order of the items was comparable to the Canadian infants. We concluded that the Canadian norms are not appropriate for the Dutch sample. Dutch infants appear to develop in a similar sequence but at a slower rate.

Like in the presented case, the AIMS is a commonly used tool to assess the current status of infant gross motor development, combined with other (observational and parental) information to create a realistic image of the current motor repertoire of the infant. Culturally valid norm references enable the PPT to support the decision-making process of whether an infant has a gross motor developmental delay compared to peers, or not.

With the insight that growing up in the Netherlands means your motor developmental pace may be slower than that of Canadian infants, we wanted to get an overview of child and environmental factors that influence gross motor development of healthy infants from birth until reaching the milestone of independent walking, based on longitudinal research. In a systematic review **(Chapter 3)**, we searched several databases (Scopus, PsycINFO, MEDLINE, and CINAHL), extracted data, and assessed the risk of bias in the selected studies, using the Quality in Prognostic Studies tool (QUIPS). We found 36 studies considering six infant and eleven environmental factors. Of these studies, five studies had a low risk of bias. Only for the factor BW, we found strong evidence for an association with gross motor development in healthy term-born and preterm born infants. There was moderate evidence for the factors GA and sleeping position. For the factors of twinning and breastfeeding we found conflicting evidence, and no evidence for the factor maternal postpartum depression with gross motor development. For the other eleven factors, including cultural influence, the evidence was limited because each of these factors was examined in only one longitudinal study. We concluded that lower BW and shorter GA have a persisting negative association with gross motor development. For many other factors, the association remains unclear, and more research is needed.

The PPT in the presented case used information about factors influencing gross motor development of premature infants to determine the infant's risk of gross motor developmental delay. Knowing that premature infants with low BW, a low five-minute Apgar score, and parents with Dutch mother tongue are factors associated with developmental delay, made the PPT even more alert.

Because motor development is one of the first signals to identify whether an infant is developing well, we wanted to explore gross motor developmental trajectories of VPT infants without severe perinatal complications. Besides, we examined whether profiles of gross motor development could be distinguished and related to comparable profiles of a sample of full-term (FT) Dutch infants **(Chapter 4)**. Parents used the AIMS home-video method to record their infant from 3-7 months CA with approximately a two-months interval until the age of 18 months CA. Forty-two Dutch infants born with ≤32 weeks GA or BW ≤ 1500 grams and their parents participated. We found also for this sample a unidirectional growth and variability in gross motor development. A trend was seen for a model in which lower BW (β= -4.10, $\beta 2$ =0 .004; p = 0.031), five-minute Apgar score <7 (β= -3.54; p = 0.033), and Dutch native speaking parents (β=-3.16; p = 0.059) were associated with slower gross motor development. Using cluster analysis, we distinguished three motor developmental profiles: early developers, gradual developers, and late bloomers. Until the age of 12 months CA, the VPT profiles were comparable to Dutch FT profiles. From 12 months CA onwards, the VPT gradual developers and late bloomers showed a delayed acceleration in the curves compared to the TB developmental profiles. For the early developers, there was no difference in the developmental curve between the VPT and TB infants. We concluded that distinguishing gross motor developmental profiles may contribute to clinical decision-making, shaping early interventions, and supporting realistic parental expectations.

As was presented in the case, the infant had a very low score at the first assessment with the AIMS. The PPT knew that only one assessment is not reliable and because motor development is very variable, she monitored the gross motor development of the infant closely.

Because VPT infants are at risk of (gross motor) developmental problems, they and their parents return to the hospital for regular check-ups at the follow-up clinic. During a check-up, standardized tests -like the AIMS- are usually administered. The purpose of the next study was to gain an understanding of parental experiences with the AIMS home-video method and we asked parents of VPT infants how they appraised its applicability for use in an outpatient follow-up clinic **(Chapter 5)**. With a qualitative research design, we interviewed 10 parents who already participated in the GODIVA-PIT study. We found that parents appraised the AIMS home-video method as manageable and fun to do. Only some practical aspects of the method, like uploading the video and recording with two people, were perceived as most bothersome. However, parents gained a better awareness of their infant's motor development and the provided feedback appeared confirmation of what they already thought. They felt reassured that their child was doing well. We also found that first-time parents seemed more uncertain and had a greater need for information about (motor) development, but on the other hand, also had confidence in their child. Lastly, all parents were of the opinion that home-videos can be an addition to follow-up visits but cannot replace (all) visits. We, therefore, concluded that parents appraised the AIMS home-video positively and that innovations with a secure and safe digital platform should be developed and implemented in neonatal follow-up, which should be explored in future studies.

The presented case happened during the COVID-19 pandemic, which made using the AIMS home-video method a useful addition, or in this case perhaps a replacement, for the PPT visits. This PPT shows that, in line with the Dutch national professional competency profile, a PPT is able to contribute to professional innovation.

Considering the stressful start parents encounter with the premature birth of their infant, with all uncertainties about future developmental problems, we were interested in parental beliefs regarding motor development and how parents consider their own role in stimulating their infant's gross motor development **(Chapter 6)**. We compared parental beliefs of parents of VPT infants with parents of FT infants (GODIVA-KIT study). All parents (37 parents of VPT infants and 110 parents of FT infants) filled out the parental beliefs (PB-MD) questionnaire when their infant was approximately 3 to 7 months old. Parents of VPT infants believed they should stimulate motor development more than parents of FT infants, but both parents tend to disagree with the belief that parents should stimulate motor development. All parents, both VPT parents as well as FT parents, think that motor development is one of the most important things during the first year of life, thought they do not hold clear beliefs in favour of actively promoting motor development. Most parents believe that they have a role in stimulating motor development, whereas parents of VPT infants more often express that they stimulate their infant by creating the right environment and/or using toys or equipment. This is important knowledge for PPTs to be able to adapt to the needs of parents in applying interventions.

The PPT in the presented case was able to meet the parental needs. Parents believed that each infant develops at its own pace but realized that their infant would benefit from more stimulation to support its motor development. Acknowledging the parental beliefs and providing information to parents, resulted in a good partnership that was beneficial for the infant.

We concluded that the PB-MD questionnaire seems not to be very sensitive to differences within the same culture and with that, it is questionable if it is applicable for clinical use. Therefore, tools for clinical use to identify parental beliefs regarding gross motor development should be developed.

In short, this thesis addressed variation in (preterm) motor development, factors of influence on (premature) infant motor development, and lastly parental beliefs and practices. Gaining insight into these elements will hopefully support early detection of gross motor developmental delay and clinical decision-making.

General Discussion

In this part of the thesis, we will discuss what the impact of our studies might be considering theoretical and methodological aspects, and which recommendations we have for future research and clinical practice, based on the reflections of the conducted research.

Theoretical construct underlying motor development

The underlying theoretical construct we had in mind when designing our studies, i.e., the grant application from which this thesis originates, was the Dynamic Systems Theory (DST). We felt this to be a suitable departure point given the DST focus on the interaction between the infant, the task and its environment as being essential for development [1,2]. In our research the infant was situated in its own (home) environment during assessments. The influence of the environment on the motor behaviour of the infant was evident. For instance, during Christmas time the infant curiously crept toward the Christmas tree in the living room, reaching to touch a Christmas figure hanging in the tree. Due to changes in the surrounding, e.g., the Christmas tree, the infant showed other, and perhaps new motor behaviour. The physical surrounding challenges the infant to experience its abilities, pushing its own boundaries to master new abilities. The interaction between the systems within the infant and its environment is a prerequisite in getting to the next step [1,2]. Time is very important in this respect. The in-the-moment behaviours have consequences for future behaviours, with the infant learning by doing and repeating, and therefore developing as time continues. We saw this happening while assessing the recordings sent by parents. Parents put their infant into prone position (task) and saw for the first time that their infant lifted its head and made eye-contact in this position. Parents were sometimes surprised, and we heard them cheering (environment) at their infant that it was doing well. The infant (child) often lifted its head again. In these recordings, we saw many infants learn and in reaction to their social surroundings, repeat what they had just learned [2,14].

Next to this being an example of the interaction of the child, the task and the environment resulting in new motor behaviour, this is also an example of a so-called *'developmental cascade'*. According to Masten and Ciccheti, developmental cascades refer to the cumulative consequences for the development of the many interactions and transactions occurring in developing systems that result in spreading effects across levels, among domains at the same level, and across different systems or generations [3]. This conceptual framework provides explanations as to why development in one domain affects development in another developmental domain, like the cognitive domain or the social domain [4–6]. In this thesis we have not investigated the interaction between motor development and other developmental domains, like the interaction between parents and infants when motor abilities increase. Though, we have obtained a considerable amount of visual and auditive data which will enable us to answer new research questions related to other developmental domains in the future. Importantly, with the DST and developmental cascades in mind, clinical decisions are based not solely on the outcome of standardized assessment(s), but also on factors influencing gross motor development, like information from parents -e.g., infant and parental factors-, clinical observations, knowledge, and clinical experience [7–9].

Measuring infant motor development

The Alberta Infant Motor Scale (AIMS)

The AIMS as a rather easy to use instrument allows clinicians to objectify gross motor development. When combined with contextual information it provides a valuable impression of the motor abilities of infants at that moment. The 58 items of the AIMS representing a motor milestone that is characterized by qualitative aspects, are either observed or not observed by the assessor. Therein lies a vulnerability: the qualitative aspects are more susceptible to interpretation. Moreover, the windows determined to represent the infants' current motor repertoire are also open to interpretation. This makes training clinicians who use the AIMS as an assessment tool important. In our studies, we countered this by training the researchers and making a priori agreements on scoring. During the study, difficult or questionable items were instantly discussed by the researcher and fellow researcher (MB[a]). In addition, the researchers independently assessed recordings that were compared. The agreement on item level between the two observers on eight infants was 97.8%.

Even though the AIMS does involve qualitative aspects of gross motor development, not all aspects of the quality of movement -e.g., fluency, and variability- are taken into account. Differences in quality of movement may be of great importance, because they may reflect and explain differences between the VPT and FT infants, which we did not capture in our study.

Reflecting on the assessments in the longitudinal study, we can be critical if the assessment at 3.5 months CA was reliable. At this age, no difference was found in average AIMS scores for VPT infants compared to FT infants **(Chapter 4)**. This is also recognized in research on the sensitivity and specificity by the developers of the AIMS where assessments before the 4th month are less sensitive and they advise using a different cut-off point (10th percentile) for identifying developmental delay [10]. The AIMS appeared to have a floor effect in the first four months after birth because relatively few items can be scored.

The AIMS home-video method

Using the AIMS home-video method made data collection easier, as it was independent of time and place. This was confirmed in the interviews with parents about the use of the AIMS home-video method. Parents appreciated the opportunity to choose the right time to record the assessment and to record their infant in its own environment **(Chapter 5)**. For some parents, it was even a prerequisite for participating in the study. Despite the advantages, also some practical disadvantages were experienced by parents, like filming with two people when the infant is young and filming on one day. For research purposes, we decided to ask parents to record the assessment on one day, because the AIMS already measures differences in scores after a week [11].

On the other hand, the AIMS home-video method also made it possible to collect data on infants from over the whole country. This contributed to a fairly representative sample of infants from different parts of the Netherlands for the AIMS-NL study **(Chapter 2)**. On the other hand, our sample did not include a representative proportion of infants of non-Western origin, as it only comprised half of the percentage of infants we should have included.

Taking into account all strengths and limitations of the AIMS and the AIMS home-video method, we think that using the AIMS as a measurement for objectifying (preterm) infant gross motor development is a valid and reliable way of collecting data for research purposes.

Part I: Factors associated with gross motor development

Looking back, and reflecting on, factors associated with gross motor development the overall picture that emerged from the literature **(Chapter 3)** and the data presented in this thesis **(Chapters 4, 5 and 6)** might be captured best by a combination and adaptation of two existing models **(see Figure 1)** that we describe next.

First, as a basis, the Developmental Niche model provides a framework in which many of the factors that are or are presumed to be associated with infant gross motor development can be placed [12,13]. Subsequently, and depicted in **Figure 1**, two elements

-moderators and developmental outcomes- of the biocultural model from Worthman were added to the Developmental Niche model [13]. More specifically, though most of the factors emerging from previous research can be placed in the Developmental Niche framework, parental characteristics like parental education, maternal depression, and maternal age do not fit in this framework. This is where the element moderators for the caretaker, proposed by Worthman, might be regarded as a valuable addition [13]. Secondly, we added infant developmental outcomes, which is the second element from the model of Worthman, because gross motor development was the major outcome measure of our research. In sum, **Figure 1** shows all the elements of our study and the interaction of the different systems concerning the infant in its (cultural) environment.

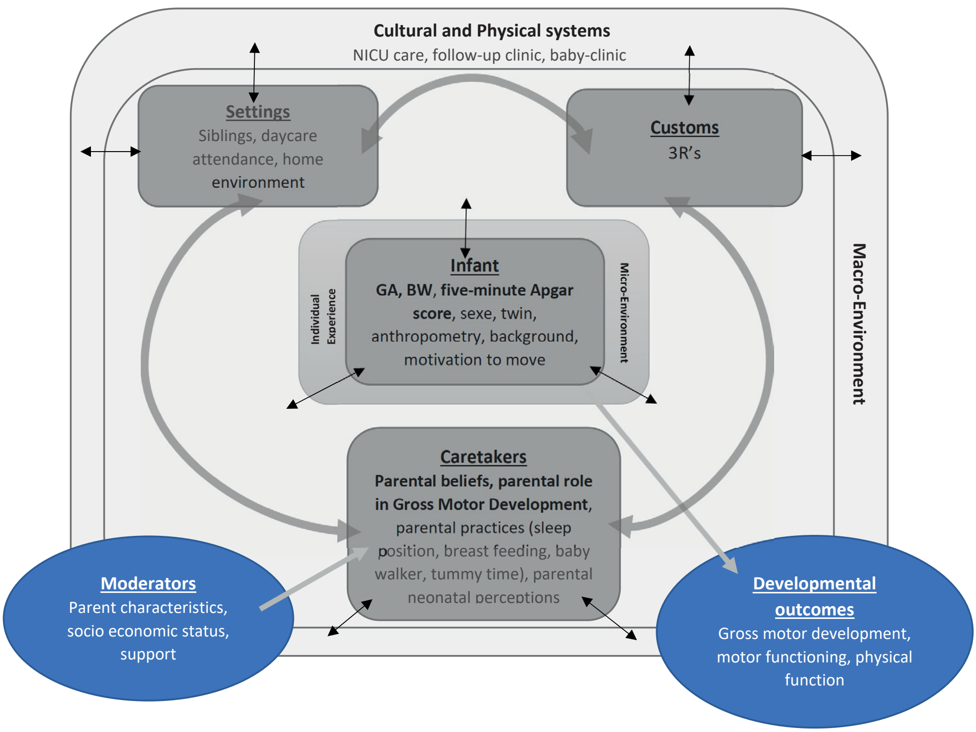


Figure 1: *Adapted from the Developmental Niche model of Harkness and Super with the two added elements -moderators and developmental outcome- of the biocultural model of Worthman [12,13].*

Cultural influences on gross motor development

According to the results of the AIMS-NL study, the Canadian norm references did not seem to be appropriate for our Dutch infants **(Chapter 2)**. Therefore, Dutch norm references needed to be established. The sample of our study was, however, too small and the level of representativeness for the Dutch population concerning infants' origin was not sufficient to establish Dutch norm references. Meanwhile, Dutch norm references have been developed [14]. Despite these new references, their sensitivity in discriminating infants with gross motor developmental delay is questioned [15].

Reasons for the differences in sensitivity compared to the Canadian norm references are still to be investigated. Explanations may be related to the composition of the sample, data collection -as data were derived from a study using another measurement tool and methodology to answer a different research question- and/or the analysis of the data.

With the cross-cultural differences in mind, and reflecting on our research, we should realize that our research was WEIRD. WEIRD stands for research among Western, educated, industrialized, rich and democratic populations. With this thesis, we try to address cultural differences being of utmost importance in gross motor development research, but also in other research domains. We perform research from our WEIRD perspective, among our WEIRD population, with our WEIRD (funding) possibilities. But, as in other literature is already emphasized and what fits with our beliefs and research findings: different cultures are not only literally a separate world, but also a different world [16–19]. The theories we develop in our WEIRD way, may not correspond to the non-WEIRD way. We should keep this in mind every time we perform our research and most of all, we should be sensitive to different or other ways of looking at research, research questions, research designs, theoretical constructs, analysis, and so on.

Part II: Infants

As we concluded in our systematic review **(Chapter 3)**, there are few infant factors with a high level of evidence for association with gross motor development. Only preterm birth is a factor within infant motor development, with a shorter GA and lower BW associated with more delayed gross motor development. This is also seen in the longitudinal study of the VPT infants **(Chapter 4)**, where three developmental profiles -early developers, gradual developers, and late bloomers- for the VPT infants were found and compared to the comparable profiles of the FT infants. The developmental profiles show that also within the group of VPT infants there is variation concerning the speed of gross motor development. From 12 months onwards, VPT gradual developers and VPT late bloomers deviated from comparable FT profiles. VPT infants' developmental pace decreased compared to the FT infants. This difference may imply that when a VPT infant has to master a more complex motor skill, it takes longer for the VPT infant to acquire this skill than for a TB infant. We only detected these differences because of the longitudinal research design with rather small age intervals. And with cluster analysis it was possible to distinguish different gross motor developmental profiles, which is also shown in other studies [20,21]. The identification of three clusters, however, is not fixed and may be different when analysing a larger research sample. Due to the small clusters, with one cluster consisting of only five infants, it was not possible to draw firm conclusions and investigate which characteristics explain the variation in developmental profiles. However, the results of this study underline the importance of multiple assessments, to capture intra-individual differences over time. But it also raises questions about timing in research designs, like where in the curve is the onset of this diversion. And could this diversion be related to sensitive periods, which are possibly different for VPT infants than for TB infants, and even perhaps different for infants in the developmental profiles [22]. To identify these sensitive periods more small measuring intervals may be needed.

Part III: Parents

The reason why cross-cultural differences in norm references of gross motor development are found is still very interesting and not simple to clarify. Which factors contribute to these differences? From the Developmental Niche perspective, differences occur due to different parental beliefs and with that, different parental practices. Childrearing practices differ between cultures, also even historically within cultures -e.g., back to sleep campaign- and influences infant motor development [16].

A rather extreme, non-WEIRD, example is seen in Tajikistan. A traditional childrearing practice in Tajikistan is 'gahvora cradling'. Infants are bound on their backs with their arms and legs constrained from moving, for up to 15 hours per day during the first 20 months of life [17]. These infants show a delay in gross motor skill achievement relative to the World Health Organisation (WHO) standards, but all (healthy) infants eventually achieve the milestone of walking and show no long-term deficits [18]. In the Netherlands, we use the playpen a lot, which in Australia is regarded as a baby-jail. We also have a fairly firm paradigm, which we call the 3Rs, 'Rust' (rest), 'Reinheid' (cleanliness) and 'Regelmaat' (regularity). This determines to a large extent how parents schedule the day with their child when it is still young [23]. Every three to four hours, the child is fed, changed, played with, and put to sleep. When a parent is cooking, the child is put in the playpen, for example, where it can amuse / play / entertain itself with a few toys. This raises the question of whether we should use standards like the WHO provides or use cultural norms [16]. And what should these cultural norms look like, considering the continuous change of diversity in the population and changes in policies, expectations, and beliefs? And should we look also at sensitive periods where life experiences can have a greater impact on development: when do these sensitive periods occur and which factors are contributing positively or negatively to these periods [22]. Perhaps these windows are less or even more culturally specific and will contribute to understanding infant development.

In our study of parental beliefs, we used the PB-MD questionnaire which was developed to measure differences in parental beliefs concerning their child's motor development between cultures. This is perhaps the reason why we did not find many differences between the parental beliefs of Dutch VPT and FT parents. Despite the few differences, we think that there are differences in beliefs among Dutch parents, and perhaps also between mothers and fathers. We are only lacking tools to gain insight or measure parental beliefs regarding motor development within our Dutch population. With such tools, we can also investigate whether parental beliefs influence infant motor development and how. And how can we also investigate differences between parents of infants with different diseases which impact motor development. These tools are also interesting to develop for different cultures, because of the different (medical) possibilities and beliefs regarding the disease.

Future research

With the case illustrating daily practice and the results of our study, obviously as befits a true researcher-, more research is needed.

- First of all, Dutch PPTs need clarification about which norm references of the AIMS to use, the Dutch norm references, or still the Canadian norm references. Perhaps the cut-off points for the new Dutch norm references need to be reconsidered, though this needs to be substantiated by research. By using accurate norm references, clinical decision-making will be greatly facilitated, and parents will get a more realistic image of their infant's gross motor development.
- Because infants are dependent on their parents to a great extent in the first period after birth, it is necessary to gain a better understanding of these parental practices. And, as mentioned earlier, it is necessary to include the quality of movement in gross motor development. Both these aims can be investigated when research is

carried out using new research methods and measurements. For example, by using video analysis [24], movement trackers [25], smart baby suits [26] and/or Ecological Momentary Assessment (EMA) [27]. To measure motor development, research must be longitudinal with short time intervals. This makes it possible to detect differences in the developmental process. And perhaps sensitive periods in development can be distinguished. When developing tools, it is very important to involve all stakeholders in the development process to ensure that all user requirements are met.

• As we experienced in the longitudinal study, clustering data seems to be one way of distinguishing different developmental trajectories in infant motor development. Generally speaking, large samples give more statistical possibilities and with that reliable results that can be generalized. However, there will always be challenges to involving large groups in research, especially those groups who already hardly participate. Other possibilities in looking at small samples should be explored, like n=1 studies and Bayesian statistics [28,29], next to already available (video)data which is open for other research questions to be answered with [30].

• The PB-MD is appropriate for cross-cultural research, but the sensitivity of the PB-MD in only the Dutch context can be questioned. Therefore, to gain more understanding of parental beliefs, what the origin of some beliefs are, whether parental beliefs change over time, their influence on infants' gross motor development, and how a PPT can acknowledge parental beliefs in daily practice, needs to be explored. It is recommended to develop tools for identifying parental beliefs for clinical and research purposes, in co-creation with PPTs, parents as well as researchers [31–33].

In all, future research should have the intention to contribute to the early detection of developmental delay, contribute to clinical decision-making, and support the development and investigation of professional and parental practices to support (preterm) infant gross motor development from birth until independent walking.

Implications for the paediatric physiotherapist

What do all these results and considerations mean for the clinical practice as presented in the case in this thesis (see General introduction and the start of this General discussion)?

For Dutch (VPT) infants, it is now evident that Canadian norm references are not appropriate. Dutch infants show a similar sequence of gross motor development, but at a slower pace. However, the recently published Dutch norm references of the AIMS also raise questions regarding sensitivity [11,12]. Consequently, using the 5th percentile as the cut-off point for the Canadian norm references would imply an overrepresentation of infants with gross motor developmental delay, and using Dutch norm references an underrepresentation of infants with a gross motor developmental delay. Of course, the PPT can still monitor progress in gross motor development of the (VPT) infant by means of multiple assessments using the raw scores. But when using the norm references, there is a risk that the current cut-off points of the AIMS may not match the clinician's and/or parent's concerns. The PPT should keep this in mind and involve parents in the shared decision-making process based on the results of all available assessments combined with the family's concerns, medical history, clinical tests, and observations. Nevertheless, for now, the dilemma of the AIMS norm references remains in clinical practice, warranting further research on cut-off points.

The factors associated with gross motor development described and identified in this thesis might aid PPTs in analysing future cases in which premature infants with a delay in gross motor development are presented to them. Moreover, the AIMS home-video method may be a valuable tool that makes it possible for PPTs to continue monitoring an infant when visits are not possible, like during the period of the COVID-19 pandemic. In addition, the method can be used to make parents more aware of their infant's gross motor development.

Furthermore, we recommend PPTs to consider and acknowledge parental beliefs when guiding infants and parents. Parents may believe that their infant will develop at its own pace, while this infant may benefit from an intervention that stimulates gross motor development. Or, when the infant is indeed doing well and monitoring the infant seems to be enough, parents can be reassured that they do not have to stimulate their infant additionally. Furthermore, we want to make PPTs aware of cultural differences and that the influence of culture is very broad and needs to be considered as a factor in the whole clinical decision-making process.

Ideally, all of the above-mentioned professional recommendations should already be taken into account during the education of PPTs. In the Dutch (3-year) master programmes for paediatric physiotherapy, it is important to give attention to the young infant in its context. The collaboration with parents deserves a larger role, by making parental beliefs a (larger) part of the curriculum. Acknowledging parents' beliefs during an intervention asks for a different attitude and not a different way of working.

Also, in the current Dutch professional competence profile so far placing the infant in its context and considering parental beliefs are not yet sufficiently explicit. We recommend the professional association, Nederlandse Vereniging voor Kinderfysiotherapeuten (Dutch Association for Paediatric Physiotherapists), to include this in a future professional competence profile.

Conclusion

We conclude that the AIMS is a robust measure for PPTs to assess gross motor development, but an answer should be given to the question of which norm references and which cut-off point should be used for identifying children at risk of gross motor problems. Besides, the AIMS home-video method seems a promising innovation that needs further development for clinical use, also for VPT infants and follow-up clinics.
Although Dutch VPT infants in general show a slower pace in gross motor development measured with the AIMS, developmental profiles reveal that a small number of VPT infants develop at a similar pace as FT infants. This emphasizes the importance of monitoring VPT infants because the larger proportion of infants develops at a slower pace from 12 months onwards.

Parents, as the primary caregivers, are the key elements for a PPT to influence gross motor development and therefore parental practices in combination with parental beliefs regarding infant gross motor development should be considered in practice and explored further.

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APPENDIX

Samenvatting

List of abbreviations

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Dankwoord

About the author

Samenvatting

Achtergrond

In Nederland worden jaarlijks ongeveer 6,7% baby's van de 164.000 baby's te vroeg geboren, wat betekent dat zij met een zwangerschapsduur van minder dan 37 weken geboren worden. Hiervan wordt 1,1% zeer vroeggeboren met een zwangerschapsduur van minder dan 32 weken. De overlevingskans van deze baby's is de laatste jaren enorm toegenomen door de verbeterde medische technologieën en zorg. Daarbij zijn nieuwe uitdagingen ontstaan in het voorkomen, diagnosticeren en behandelen van mogelijk nadelige gevolgen van vroeggeboorte op de ontwikkeling van het kind. Zo worden, bijvoorbeeld, bij 20-36% van deze kinderen nog steeds op lagere schoolleeftijd problemen met grove en fijne motoriek, cognitie en/of gedrag gerapporteerd.

Een zeer vroeggeboren baby wordt opgenomen op de Neonatale Intensieve Care Unit (NICU). Na een, vaak langdurige, ziekenhuisopname mag de baby met de ouders mee naar huis. Vanaf dat moment wordt de ontwikkeling van de baby goed in de gaten gehouden en worden ouders uitgenodigd om met hun kind regelmatig terug te komen op de polikliniek neonatologie voor follow-up. Tijdens deze bezoeken wordt de neurologische, motorische, cognitieve en gedragsontwikkeling geobserveerd en beoordeeld met het achterliggende idee dat vroegtijdige opsporing van ontwikkelingsproblemen vroege interventies mogelijk maakt.

De vroeg grofmotorische ontwikkeling is bij uitstek één van de gebieden die zich leent een beeld te vormen van het ontwikkelingsverloop van het kind en daarmee een goede indicator voor een mogelijke ontwikkelingsachterstand. Er is echter veel variatie in het verloop van de grofmotorische ontwikkeling. Dat maakt het herkennen van vertraagde motorische ontwikkeling moeilijker. Het herhaaldelijk meten van de grofmotorische ontwikkeling op jonge leeftijd maakt het voor een kinderfysiotherapeut mogelijk zicht te krijgen op de werkelijke motorische ontwikkeling en zo nodig tijdig een interventie te starten. Om de grofmotorische ontwikkeling in kaart te brengen, wordt in Nederland (en andere landen in de wereld) de Alberta Infant Motor Scale (AIMS) vaak gebruikt als meetinstrument. Naast het observeren en beoordelen van de motoriek maakt de kinderfysiotherapeut een inschatting van (andere) kind- en omgevingsfactoren mogelijk van invloed op de grofmotorische ontwikkeling. Wanneer uit deze analyse blijkt dat interventie nodig is, is een goede samenwerking met ouders belangrijk om de beste zorg te bieden voor hun kind.

Doel van het proefschrift
De Introductie (**hoofdstuk 1**) van het proefschrift beschrijft het doel, welke drieledig is: 1) het in kaart brengen van factoren van invloed op de grofmotorische ontwikkeling bij (vroeggeboren) baby's, 2) het onderzoeken van de ontwikkeling van de grove motoriek van zeer vroeggeboren baby's en 3) het onderzoeken van de ervaringen met het afnemen van de AIMS middels een home-video methode en het in kaart brengen van de gedachten en overtuigingen van ouders ('parental beliefs') over de motorische ontwikkeling van hun kind.

Deel I: Kind- en omgevingsfactoren

In **hoofdstuk 2** is onderzocht of de momenteel gebruikte Canadese normen van de AIMS geschikt zijn voor baby's in Nederland. Met de AIMS home-video methode hebben 499 ouders van typisch ontwikkelende baby's, in de leeftijd van 2 weken tot 19 maanden, hun baby éénmalig gefilmd. Uit de resultaten bleek dat 45 van de 58 items van de AIMS geschikt waren voor analyse. Van deze 45 items werden 42 items door Nederlandse baby's op een latere leeftijd behaald in vergelijking tot de Canadese normgroep. De volgorde van het behalen van de items van de AIMS was vergelijkbaar met die van de Canadese baby's. Wij concluderen dat de huidige Canadese normen niet geschikt zijn voor de Nederlandse steekproef. Nederlandse baby's lijken zich in een vergelijkbare volgorde te ontwikkelen, echter in een langzamer tempo.

Met het inzicht dat er verschillen bestaan in motorisch ontwikkelingstempo tussen kinderen die opgroeien in Nederland en Canada, werd een longitudinaal onderzoek gestart naar kind- en omgevingsfactoren die de grofmotorische ontwikkeling van gezonde (à terme of premature) baby's beïnvloeden vanaf de geboorte tot het bereiken van de mijlpaal van zelfstandig lopen (**hoofdstuk 3**). In een systematische review, exclusief gericht op longitudinale studies, doorzochten we verschillende databases, extraheerden data en beoordeelden de kwaliteit van 36 geïncludeerde artikelen met de Quality in Prognostic Studies (QUIPS) tool. Er werden veel factoren gevonden geassocieerd met de vroege grofmotorische ontwikkeling, maar slechts weinig factoren die in meerdere (longitudinale) studies onderzocht zijn. Wij concluderen uit deze review dat een lager geboortegewicht en een kortere zwangerschapsduur een blijvend negatief verband hebben met de grofmotorische ontwikkeling. Voor veel andere factoren blijft het verband onduidelijk en is meer onderzoek nodig.

Deel II: Baby's

Omdat de motorische ontwikkeling één van de eerste signalen is om vast te stellen of een baby zich goed ontwikkelt, werden de grofmotorische ontwikkelingstrajecten van zeer vroeggeboren baby's zonder ernstige perinatale complicaties gemodelleerd (**hoofdstuk 4**). Daarnaast onderzochten we of profielen van grofmotorische ontwikkeling onder zeer vroeggeboren kinderen (d.w.z. groepen van kinderen met een vergelijkbaar ontwikkelingstempo) konden worden onderscheiden en de mate waarin deze overeenkomen met profielen van een steekproef van voldragen Nederlandse baby's. Ouders van zeer vroeggeboren kinderen gebruikten de AIMS home-video methode om de grofmotorische ontwikkeling van hun baby te registreren vanaf 3-7 maanden gecorrigeerde leeftijd, met ongeveer twee maanden interval, tot de leeftijd van 18 maanden. Tweeënveertig ouders van baby's geboren ≤32 weken zwangerschapsduur of ≤ 1500 gram geboortegewicht, namen deel. Er werd een trend gezien voor een model waarin een lager geboortegewicht, een Apgar-score op 5 minuten lager dan 7 en een Nederlandstalige ouder geassocieerd waren met een tragere grofmotorische ontwikkeling. Er werden drie profielen gevonden; vroege ontwikkelaars, geleidelijke ontwikkelaars en laatbloeiers. Voor alle drie de profielen gold dat tot 12 maanden (gecorrigeerde) leeftijd een gelijke grofmotorische ontwikkeling te zien was met vergelijkbare profielen van op tijd geboren baby's. De geleidelijke ontwikkelaars en laatbloeiers buigen na 12 maanden af naar beneden. Opvallend was dat het profiel van de vroege ontwikkelaars ten opzichte van de op tijd geboren baby's een vergelijkbare grof motorische ontwikkeling lieten zien tot aan het loslopen. Wij concluderen dat het onderscheiden van grofmotorische ontwikkelingsprofielen kan bijdragen aan klinische besluitvorming, het vormgeven van vroege interventies en het ondersteunen van realistische verwachtingen van ouders.

Deel III: Ouders

Omdat zeer vroeggeboren baby's een risico lopen op (grofmotorische) ontwikkelingsproblemen, komen zij en hun ouders regelmatig terug naar het ziekenhuis voor controles op de follow-up kliniek. Tijdens een controle worden meestal gestandaardiseerde tests -zoals de AIMS- afgenomen. Het doel van de volgende studie (**hoofdstuk 5**) was om inzicht te krijgen in de ervaringen van ouders van zeer vroeggeboren kinderen met de AIMS home-video methode en hoe zij de toepasbaarheid inschatten van deze methode voor gebruik in een poliklinische follow-up polikliniek. Met een kwalitatieve onderzoeksopzet werden 10 ouders geïnterviewd. Resultaten lieten zien dat ouders de AIMS home-video methode beoordeelden als hanteerbaar en leuk om te doen.

Alleen enkele praktische aspecten van de methode, zoals het uploaden van de video en het opnemen met twee personen, werden als lastig ervaren. Ouders kregen door het filmen van hun kind en de daarop ontvangen feedback van een kinderfysiotherapeut een beter inzicht in de motorische ontwikkeling van hun kind. Bovendien bleek de gegeven feedback een bevestiging van wat ouders al dachten over de motorische ontwikkeling van hun kind: ze voelden zich gerustgesteld dat hun kind het (over het algemeen) goed deed. We vonden verder dat ouders voor wie het deelnemende kind aan de studie hun eerstgeborene was, onzekerder leken en meer behoefte hadden aan informatie over de (motorische) ontwikkeling, maar anderzijds ook vertrouwen hadden in hun kind. Ten slotte waren alle ouders van mening dat thuisvideo's een aanvulling kunnen zijn op vervolgbezoeken in het ziekenhuis, maar niet (alle) bezoeken kunnen vervangen. Wij concludeerden daarom dat ouders van zeer vroeggeboren kinderen de AIMS home-video positief beoordeelden en dat innovaties met een veilig digitaal platform moeten worden ontwikkeld en geïmplementeerd in de neonatale follow-up, wat in toekomstige studies moet worden onderzocht.

De laatste studie (**hoofdstuk 6**) betrof het inzicht krijgen in de ‘beliefs’ - ideeën, gedachten en overtuigingen - van ouders van een zeer vroeggeboren baby. Gezien de stressvolle start die ouders ervaren bij de vroeggeboorte van hun kind, met alle onzekerheden over toekomstige ontwikkelingsproblemen, waren wij geïnteresseerd in de ‘parental beliefs’ met betrekking tot de motorische ontwikkeling van hun kind en hoe ouders hun eigen rol zien in het stimuleren van de grofmotorische ontwikkeling van hun kind. Wij vergeleken de ‘parental beliefs’ van 37 ouders van zeer vroeggeboren baby’s met die van 110 ouders van op tijd geboren baby’s middels hun antwoorden op de Parental Beliefs on Motor Development (PB-MD) vragenlijst ingevuld toen hun baby 3 tot 7 maanden (gecorrigeerde leeftijd) oud was. Zowel de ouders van de zeer vroeggeboren baby’s als van de op tijd geboren baby’s neigden meer naar de overtuiging dat het stimuleren van de motorische overtuiging niet nodig is. Ondanks dat was er toch een significant verschil tussen de groepen ouders, waarbij ouders van de zeer vroeggeboren baby’s het meer eens zijn met de opvatting dat ouders hun kind moeten stimuleren. Alle ouders, zowel ouders van zeer vroeggeboren baby’s als ouders van op tijd geboren baby’s, vonden de motorische ontwikkeling één van de belangrijkste zaken tijdens het eerste levensjaar. De meeste ouders vonden dat zij wel een rol hebben in het stimuleren van de motorische ontwikkeling, waarbij ouders van zeer vroeggeboren baby’s vaker aangaven dat zij hun baby stimuleren door de juiste omgeving te creëren en/of speelgoed te gebruiken in plaats van actief stimuleren van de motorische ontwikkeling. Dit is belangrijke kennis voor kinderfysiotherapeuten om, bij het toepassen van interventies, te kunnen inspelen op de behoeften van ouders.

In **hoofdstuk 7** worden de resultaten van de uitgevoerde studies die deel uitmaken van dit proefschrift samengevat en een reflectie gegeven op de (hoofd)uitkomsten. Aan de hand van de Dynamische Systeem Theorie en de Developmental Niche theorie wordt teruggekeken op de dynamiek en variatie in grofmotorische ontwikkeling van (zeer vroeggeboren en op tijd geboren) kinderen inclusief factoren geassocieerd met het verloop en de snelheid van de grofmotorische ontwikkeling, waaronder de invloed van het opgroeien in de eigen culturele context. Dit laatste hoofdstuk van het proefschrift wordt afgesloten met aanbevelingen voor de klinische praktijk en toekomstig onderzoek.

List of abbreviations

ABW	Adequate Birthweight
AHEMD-IS	Affordances of the home environment - Infant-Scale
AIC	Akaike Information Criterion
AIMS	Alberta Infant Motor Scale
ASQ-II	Ages and Stages Questionnaire, second edition
BMI	Body Mass Index
BPD	Bronchopulmonary Dysplasia
BSID	Bayley Scales of Infant Development
BW	Birthweight
CA	Corrected Age for prematurity
CP	Cerebral Palsy
DAIS	Daily Activities of Infants Scale
DDST	Denver Developmental Screening Test
DST	Dynamic Systems Theory
EPDS	Edinburgh Postnatal Depression Scale
EPT	Extremely Preterm
FCC	Family Centered Care
FT	Full Term
GA	Gestational Age
GMA	General Movements Assessment
GMD	Gross Motor Development
HBW	High Birthweight
ICC	Intraclass Correlation Coefficient
IMP	Infant Motor Profile
IMQ	Infant Motor Quotient (now ASQ)
IVH	Intraventricular Hemorrhage
KIDI	Knowledge Infant Development Inventory
LBW	Low Birthweight
LMM	Linear Mixed Model
LNF	Dutch Neonatal Follow-Up (LNF) Study Group
M	Mean
M-ABC	Movement-ABC
MAI	Movement Assessment of Infants
MD	Motor Development
MLBW	Medium Low Birthweight

MM	Motor Milestones
MPT	Moderately Preterm
MSEL	Mullen Scale of Early Learning
MTM	Motivation to Move scale
N/A	Not Applicable
NBAS	Neonatal Behavioral Assessment Scale
NBW	Normal Birthweight
NEC	Necrotizing Enterocolitis
NICU	Neonatal Intensive Care Unit
NPI	Neonatal Perception Inventory
PBs	Parental Beliefs
PB-MD	Parental Beliefs on Motor Development questionnaire
PDI	Psychomotor Developmental Index
PDMS	Peabody Developmental Motor Scales
PPD	Postpartum Depression
PPMD	Postpartum Maternal Depression
PT	Preterm
PPT	Pediatric Physical Therapist / Therapy
QUIPS	Quality in Prognostic Studies tool
RDS	Respiratory Distress Syndrome
RoB	Risk of Bias
SD	Standard Deviation
SDC	Smallest Detectable Change
SEM	Standard Error of the Measurement
SES	Socioeconomic Status
SF-36	36-item Short Form Health Survey
TB	Term Born infant
TBCS	Taiwanese Birth Cohort Study developmental instrument
TD	Typically Developing
TOP	Transmural developmental support for VPT infants and their parents
VLBW	Very Low Birthweight
VPT	Very Preterm

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'Stap voor stap', dat is de titel van mijn proefschrift. Een kind ontwikkelt zich stap voor stap tot in de volwassenheid, vergelijkbaar met de totstandkoming van dit proefschrift, dat zich ook stap voor stap ontwikkeld heeft. Het was een proces waarin ik veel geleerd heb, niet alleen over alles wat ik heb mogen onderzoeken, maar ook over mijzelf als persoon.

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Curriculum Vitae

Imke Suir was born in Leiden, the Netherlands, on March 19th, 1980. In 1998, she received her VWO-diploma at the Hanfortmann College in Heerhugowaard. She completed her Movement Science study at the University of Groningen in 2006 and her Physical Therapy study at the HU University of Applied Science in Utrecht in 2007 and started working at the Tyltylschool, a school for children with a physical and intellectual disability (Den Haag). In 2007 she started with the specialization in Pediatric Physical Therapy at the Transfergroup Rotterdam, which she finished in 2011. In the meantime she worked as a Pediatric Physiotherapist in different outpatient care settings and schools. In 2013 she started as a Pediatric Physiotherapist and Junior Researcher at Rehabilitation Centre De Hoogstraat for the project @home which ended in 2015. Then Imke started at the HU University of Applied Science in Utrecht as a lecturer at the Minor Child in Movement. The same year, she started participating as a researcher in the GODIVA project (Gross mOtor Development of Infants using home Videos with the AIMS) at the HU, Research group Lifestyle and Health. In 2016, she received a personal teachers grant from the Dutch Scientific Organization (NWO) and started her PhD with the project name GODIVA-PIT (Premature Infants following in Time). The research was about gross motor development of very preterm infants and associated factors. In September 2022 Imke started participating in a new project PEBBLES (ParEntal Beliefs concerning their Baby Lifestyle and Experience Study) while finalizing her PhD and she will continue applied research into gross motor development of infants and parental beliefs, and dissemination of knowledge by lecturing in the Master Pediatric Physiotherapy.

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