Chapter 7: General Discussion
The studies reported in chapters 2-6 have increased our knowledge on the development of postural control in typically developing infants and infants at risk of cerebral palsy (CP). However, the results are not always easy to interpret. In the following discussion, I have endeavoured to interpret the results within the theoretical frameworks of the Neuronal Group Selection Theory and Dynamic Systems Theory (see also the Introduction in chapter 1). These frameworks serve as a valuable tool to place the results in a developmental context. First, some attention will be devoted to methodological considerations. Thereafter, postural control in typical development will be addressed, followed by postural control in infants at risk of cerebral palsy. The chapter concludes with possible implications for intervention.

**METHODOLOGICAL CONSIDERATIONS**

The study results from this thesis demonstrate that postural adjustments produced in a natural setting (during an internally triggered reaching movement) are very different from postural adjustments in reaction to the somewhat artificial balance disturbances provided by platform perturbations. This means that to fully grasp the complexity of postural development, one should not only study reactive postural control in laboratory situations, but also proactive postural control during natural, spontaneous motor behaviour.\(^1\) This provides a challenge to the researcher, as the timing of the movements is not precisely controlled. The newly developed PedEMG programme was designed to facilitate and improve accuracy of data analysis of spontaneous muscle recruitment, by integrating video and EMG analysis and using a dynamic statistical algorithm for onset detection in the EMG signals.\(^2\) However, even with this advantage, analysing the muscle recruitment strategies was a considerable challenge. The reason for this is the ‘noise’ of muscle activation in the infant EMG that - as I mentioned in the introduction - should actually not be regarded as noise at all, but as a characteristic of the exploratory nature of motor development (learning by trial-and-error). While the adult EMG shows a clear difference between a resting state and a purposeful muscle contraction, the infant EMG shows continuous exploratory muscle activation both between and during reaching movements. This makes it very hard to classify muscle activation as part or
not part of the reaching movement and is an important explanation for the variability we found in the data.

Infant research with EMG is challenging for another reason. While cooperation of an adult can easily be obtained by explaining the procedure, the cooperation of an infant depends on very different parameters. In the early intervention project (Dutch: Vroegtijdige interventieproject or VIP) the infants were assessed at the research lab, which for some infants was such a strange environment that it interfered with cooperation. In addition, the EMG surface electrodes were a very interesting phenomenon to many of the older infants, resulting in the infants’ exploration of these strange sticky things on their skin. Combined with some technical issues of the video and EMG recording, this unfortunately resulted in a considerable loss of data (chapters 2-4). The pattern of missing data seemed completely random, but in hindsight affected the TIP group (typical infant physiotherapy) at 18 months more than the COPCA group (COPing with and Caring for infants with special needs) at the same age. The small number of TIP infants with postural data at 18 months may have introduced bias in the results of this group at this age. We therefore included possible confounders in our statistical models to minimize bias (see chapter 3). The data from the TIP group at 18 months should nevertheless be interpreted with caution. To deal with the missing data as best as we could, we changed our statistical approach from nonparametric tests (chapters 2 and 5) to mixed models and generalized estimating equations (GEE; chapters 3, 4 and 6), which are more robust in datasets with missing data. We also learned from our mistakes in the VIP project and changed the approach to the EMG measurements to limit the missing data in the L2M 0-2 project: the EMG measurements in the latter project were mostly done with mobile equipment at the infant’s home and planned to suit the infant’s daily routine to maximize infant cooperation.

Group sizes are usually small in this type of infant research, which was also the case in the research of this thesis. Therefore we should be careful when generalizing conclusions to the general population. Results may be subject to chance, or only applicable to the group under investigation. However, we did replicate some results in two different groups of infants (both typically developing infants and infants at risk of CP), such as the increase with age in direction-specificity at the trunk level in typically developing infants, and lack of this increase in infants at risk of CP. In other
words, although groups are small, they may still increase our knowledge of the real world.

Another limitation is the heterogeneity of the infants studied, particularly the high-risk infants. It is a bit of a paradox that the very nature of the developing brain, i.e. its plasticity and adaptability, designates early infancy as a favourable time window for physiotherapeutic intervention, while at the same time making it harder to identify which children will actually need the intervention. With improved imaging techniques and general movement assessments, it is possible to identify infants at risk of cerebral palsy before the age of 6 months, yet the final diagnosis and severity of motor impairment can only be reliably established at around 2 years of age. The infants studied for the VIP-project were included based on abnormal general movements, a selection that resulted in a group of infants at high risk of CP. Although almost all infants turned out to have minor neurological dysfunction, only 25% of these infants developed CP. In the L2M 0-2 project, even higher-risk infants were selected based on either brain lesions or neurological dysfunction or both. In this group, 54% of infants were diagnosed with CP (in the subgroup of infants who learned to walk, described in chapter 6, this percentage was 18%). This illustrates that even though we can identify infants at risk, these high-risk groups are composed of neurologically diverse infants, resulting in heterogeneous groups in the analyses. However, it is still useful to study these groups, since the infants receiving early intervention in daily practice constitute similar groups based on risk factors, due to the very same inability to predict the future.

**POSTURAL CONTROL IN TYPICAL DEVELOPMENT**

**The importance of variation**

The first thing that attracts attention in all data analyses across the studies, is that infant postural muscle recruitment is characterized by variation. Variation in strategies was visible in virtually all postural parameters studied. This suggests that there are benefits to the ability to vary. Why is variation so important?

First, I will return to the definitions of variation and variability, to avoid misconceptions in the narrative below. This is important because, even when the discussion is confined to the field of motor behaviour and development, the words
variation and variability have many different connotations for different people. With variability, I mean ‘the ability to vary’ in a general sense. As a result of variability of the neural activity patterns, there are differences, or variation, in motor behaviour and motor strategies. Variability and variation of motor behaviour reflect the complex array of signalling possibilities of the neural network (the nervous system). Recall from chapter 1 that we can distinguish between a primary repertoire that is formed by connections between (groups of) neurons determined by genetic programming and epigenetic events, and a secondary repertoire that is shaped by the effect of experience on the intersynaptic connections. The secondary repertoire is the result of the combination of variation and adaptability, i.e., the ability to learn to select the best response in a given situation and thus adapt the neural activity to the specifics of the situation. Variation can be explorative in nature (exploring all the possibilities of the primary repertoire) or adaptive (as numerous adaptive strategies can be available in the secondary repertoire). The latter may be called adaptive variability. The word adaptability puts more emphasis on selection, while the term adaptive variability puts more emphasis on having several alternatives: the ability to vary. They are like the two sides of a coin.

The most obvious benefit of variability is that of learning by trial-and-error. In terms of the neuronal group selection theory (NGST), variation illustrates the trial-and-error process of trying out different strategies to find out which ones are most successful. With subsequent selection, the infants gradually learn to apply the successful strategies (adaptability). The selection of direction-specific postural adjustments with age, as seen in chapters 2 and 6, is an example of this process.

However, as detailed above, it should be noted that variation is not only characteristic of the ‘learning phase’ of skill acquisition (explorative variability), but also of using the skill thereafter (adaptive variability). This was also visible in our data, where postural strategies were characterized by variation even in the older infants, at an age when reaching for a toy in a sitting position can hardly be considered a novel task. This adaptive variability is often overlooked because in general, skilled motor performance is characterized by consistent movement patterns. In other words, skilled motor performance produces less variable motor output than novice motor performance (despite the presence of a varied repertoire in the skilled performer). This easily leads to the assumption that variation in motor performance is a ‘problem’ that should be minimized (i.e., reduced to zero). The underlying
thought is that the variation in outcome measures represents the variation in the movement patterns that produced the outcomes. However, this is not necessarily true: several different strategies can lead to the same, consistent outcome, and increasing consistency of the motor result is not the same as minimizing variation of motor performance. In fact, in terms of NGST, variability in motor behaviour enables (and thus reflects) adaptation of strategies to the specifics of the situation. For example, recruitment order of postural muscles can be top-down to facilitate hand-eye coordination, or bottom-up to facilitate trunk stability, depending on which has - at that moment – the highest priority (see chapter 2). Thus, variation in movement patterns could be interpreted as reflecting the conscious or unconscious attempt to optimize the interaction between motor behaviour and the combined constraints of the situation - a process referred to as self-organising optimality by Newell. Apart from optimizing motor performance, this may have a beneficial effect on safety, by providing the flexibility necessary to adjust to sudden alterations in the environment (such as changes of the support surface) and thus reduce the risk of injury through falling. It has even been suggested that variability in motor coordination helps to prevent repetitive strain injuries and that decreased variability of coordination in an impaired limb can be compensated by increased coordination variability of the non-impaired limb, both in adults with patellofemoral pain and in children with cerebral palsy.

Another essential function of motor variability is probably that it provides sensory information that helps solving motor problems, by externalizing (and thereby simplifying) the problem solving process. As already pointed out in the introduction, the human brain is not a logical processing machine separated from its physical environment that gets input from one system and provides output through another system. Rather, it is an integrated network that functions as a whole, and actively uses motor behaviour as a source of information in order to reduce the complexity of problem solving. This is called ‘embodied cognition’: the body and its motor behaviour are part of the problem solving process. For example, when completing a jigsaw puzzle, we actively pick up the pieces and turn them around to see if they fit rather than just looking at all the pieces and mentally turning them around, which is a computationally laborious process. The same holds for postural sway: rather than continuously predicting the effect of all the different parameters that are acting on the body (gravitational forces, inertia and so on) to determine the
best muscle contraction pattern to maintain balance, we actively explore the limits of stability, thus acquiring sensory information about the interaction between the body and the environment that actively helps maintaining balance.\textsuperscript{14,15} In terms of dynamic systems, the complex interplay between motor behaviour, sensory processing and constraints of the environment results in a dynamic exploration of possibilities, which brings about new opportunities for acquiring knowledge about the world.\textsuperscript{16} Motor variability is a functional entity that facilitates the discovery and adoption of optimal states of coordination.\textsuperscript{9} Summing up, variation of motor behaviour is beneficial for learning, adaptation and problem solving and is a hallmark of healthy motor development.\textsuperscript{7,17,18}

**Adaptability is task-specific**

At the basic level of postural control, the successful strategy in case of a balance disturbance is to apply direction-specific postural adjustments. Infants as young as one month of age already use direction-specific responses to platform perturbations.\textsuperscript{19} Consistent direction-specific adjustments are also seen during reaching movements in children from 2 years onwards.\textsuperscript{20} It may seem contradictory, then, that typically developing infants between 4 and 10 months of age use direction-specific adjustments in only half of the reaching movements (chapter 2). It may be that selecting a direction-specific strategy is easier for large balance disturbances than it is for smaller ones, or in other words that the ability to select direction-specific postural adjustments is task-specific.

In a sense, this is not surprising. After all, the phase of adaptability starts at function-specific ages.\textsuperscript{8} Certain constraints must be met, both in terms of prior experience and in terms of the development of the central nervous system, before the selection phase for a certain function can begin. Thus, the selection of adaptive strategies for head stabilization starts already around one month of age, while adaptive strategies for walking usually are not selected until around the first birthday. The process is further influenced by task constraints. For example, toddlers who have mastered walking (and thus have entered the phase of adaptive variability for walking), are not yet proficient at walking on slippery slopes (they still have difficulty selecting the optimal strategy for walking when task constraints are different).\textsuperscript{21}

In adults, it has been shown that the type and intensity of a balance disturbance alters the postural response.\textsuperscript{22} Furthermore, postural strategies have been shown in
adults to be influenced by many different variables: constraints imposed by the environment, biomechanics, body properties, forces acting on the body, support surfaces, earlier experience and practice, and the goal of the task. Therefore, one can expect the application of postural strategies to vary, both in the learning phase (during which many possibilities are explored to find out which strategies fit which situations), and in the adaptive phase (during which the postural strategy is adapted to variations in the situational constraints). This explains why even basic postural strategies, such as direction-specific adjustments, are applied differently in different situations. In infants, even small changes in task constraints can alter the chosen postural strategy: postural responses in infants are different when holding a toy compared to not holding a toy. Thus, the larger postural disturbance of the perturbation experiments may induce direction-specific postural adjustments at an earlier age than the more subtle postural disturbance due to a voluntary arm movement. In case of the latter, selecting a suboptimal strategy usually doesn’t have severe consequences, so learning the difference between optimal and suboptimal strategies requires more trial-and-error experience, and occurs at a later age. This happens at around 18 months of age, when typically developing infants show an increase in direction-specific responses in the trunk (chapters 2 and 6).

With respect to recruitment order, the picture is a little less clear. The recruitment order of the postural muscles during a reaching movement was mostly characterized by variation, but the typically developing infants did show a modest preference for top-down recruitment that gradually changed to a modest preference for bottom-up recruitment at 18 months (chapters 2, 3 and 6). These modest preferences may reflect which frame of reference is used for stabilization of the body parts. For top-down recruitment, the head functions as a reference frame, prioritizing head stabilization in space. This may be beneficial during the period in which reaching movements are developed, since it facilitates hand-eye coordination. During the period of acquisition of stance and independent walking, the reference frame may shift more to the base of support, thus resulting in more frequent bottom-up recruitment. However, the differences were small in terms of percentage of trials and variation was still abundant. It may be that selection of a specific recruitment order starts at a later age, or that this variability reflects adaptations of the postural strategy to subtle trial-to-trial differences, for example in the position of the toy, the
position of the infant at the start of the reaching movement, and the trajectory of the reaching movement.

**POSTURAL CONTROL IN INFANTS AT RISK OF CEREBRAL PALSY**

**Impaired adaptability**

In the postural parameters we studied, variation was present not only in the typically developing infants but also in the infants at risk of CP (chapters 3-6). In terms of these postural adjustments, therefore, the postural repertoire of the infants at risk of CP seemed similar to that of typically developing infants. However, since we zoomed in on a specific aspect of postural control (i.e., muscle recruitment strategies) during a specific task (reaching while sitting) and did not design the studies specifically to measure the amount of variation, these results do not exclude the possibility that infants at risk of cerebral palsy have a smaller postural repertoire than typically developing infants. There is evidence for a smaller motor repertoire in other studies. For example, infants who later develop cerebral palsy display general movements with reduced variation in early infancy,\(^27\) and older children with CP have gait patterns with reduced complexity compared to typically developing children.\(^{28,29}\) In the case of postural control during sitting, a very limited postural repertoire has been found in two children with severe CP (GMFCS level V) who never learned to sit independently.\(^{30,31}\) These children showed a total lack of direction-specific postural adjustments, displaying a complete inability to apply this basic strategy of postural control. Children with less severe forms of CP do have the ability to produce direction-specific postural adjustments, but do so less consistently and at a later age than typically developing children.\(^{32}\) Apparently, direction-specificity is part of their postural repertoire, but learning to select it at the right moment is more difficult for these children than for typically developing children. This may reflect impaired adaptability in children with CP.

In the studies in chapters 3, 4 and 6 we found a similar pattern of impaired adaptability of direction-specific postural adjustment during reaching in infants at (very) high risk of CP. While typically developing infants learned to consistently apply direction-specific postural adjustments during reaching, this selection was absent in the infants at risk of cerebral palsy; throughout infancy, they only used direction-
specific adjustments in about half the number of trials. Since children with CP eventually learn to consistently use direction-specific postural adjustments during reaching, the lower direction-specificity at 18–20 months of age in the (very) high-risk infants can be interpreted as a developmental delay. A possible explanation for this delay, as suggested in chapters 3 and 4, is impaired sensory processing in infants at risk of CP\textsuperscript{33,34}; sensation is an important factor in motor learning.\textsuperscript{35} Actually, this is not surprising: learning requires feedback, and feedback is received in the form of sensory information. If sensory processing is impaired, then trial-and-error learning is less effective and requires more experience. In other words, the number of trials needed for a similar learning result is much higher in children with CP, if a similar result is obtained at all.

Such impaired adaptability has also been observed in other parameters of postural control. Fine-tuning of the postural response can be achieved by adapting the number of direction-specific muscles that are activated, the order in which they are activated, and the strength of the contraction (visible as amplitude modulation of the EMG signal). In a study comparing children with CP to typically developing children, postural responses to repeated perturbations were examined to evaluate short-term learning on postural adjustments.\textsuperscript{36} Typically developing children quickly down-graded their postural adjustments (in terms of the number of muscles recruited and the EMG amplitude) to the minimum of activation that was needed to prevent falling. The children with CP, however, adapted their postural response at a slower pace and to a lesser degree than typically developing children.\textsuperscript{36}

In the studies described in this thesis, we did not see differences in the number of postural muscles used during the reaching movements; responses varied but the dominant pattern of activation was the complete pattern in which all three direction-specific muscles participated, for both typically developing infants and infants at risk of CP. However, we used a very sensitive method for onset detection, resulting in a high number of participating muscles even when the strength of contraction was low. There may have been differences in amplitude modulation, but we did not analyse that parameter in the current studies. We did see more antagonistic co-activation of the rectus abdominis muscle in the very high-risk infants (chapter 6), a feature that has been reported before in children with CP.\textsuperscript{37–39} We also found differences in recruitment order. Within the variation, the typically developing infants changed from a slight preference for top-down recruitment in early infancy to a slight
preference for bottom-up recruitment (chapter 2). This trend was not visible in the adjustments of infants at risk of CP. Curiously, in our at-risk infants we did not see the stereotyped top-down recruitment that was found in an earlier study in children with cerebral palsy. Perhaps this preference is only found in more severely affected children: in the VHR-infants in chapter 6, top-down recruitment was more frequent in the infants who later developed CP than in those who did not. The difference, however, was small. It may also be that stereotyped top-down recruitment during reaching movements takes more time to develop.

Possible implications for intervention

If impaired adaptability is indeed (partially) caused by impaired sensory processing and sensorimotor integration, then children with or at risk of CP might benefit from additional feedback to compensate for their own reduced information processing. Indeed, providing additional real-time feedback, for example as visual and auditory cues in virtual reality, can help children and adults with CP to improve gait parameters as well as postural parameters. An increased amount of trial-and-error experience may also help. To test this hypothesis, we compared postural adjustments of at-risk infants who had received the physiotherapy intervention programme “COPing with and CAring for infants with special needs” (COPCA) to those who had received typical infant physiotherapy (TIP; see chapter 4). We did not find significant differences in postural parameters at RCT level. However, a process evaluation of the physiotherapy the infants had received, demonstrated that the contents of the intervention were heterogeneous within the groups and partially overlapped between the groups. Relating the process evaluation to postural adjustments suggested that intervention that includes spontaneous motor behaviour with trial-and-error and without interference of the therapist is associated with improved postural development (chapter 4). Therefore, although the effect sizes are probably small, stimulating self-produced motor behaviour (and thus increasing trial-and-error experience) is likely to benefit postural development. It is important to let the child not only experience the ‘trial’-component, but also the ‘error’: all too often, we try to impose the ‘right’ strategy, depriving the child of the opportunity to explore and thus learn from his/her mistakes.

Strikingly, more than 50 years ago Myrtle McGraw had already come to a similar conclusion in her description of the studies with the identical twins Jimmy and
Johnny. By teaching Johnny new skills while leaving Jimmy to grow up without any intervention, she showed that practice could advance the acquisition of certain motor skills, and induce changes in motor performance that were measurable later in life. In these experiments, she tried to teach Johnny to ride a tricycle at approximately 12 months of age, by putting him on the tricycle with his feet tied to the pedals, twice a day, five days a week, for 7 months. With this experimental set-up, the only two options for the child were to perform the complex action of tricycling the right way, or not at all. At the age of 19 months, the child finally discovered the right strategy. Later, McGraw wrote: “Now, in retrospect, I’m quite confident that if we had not tied that baby’s feet to the pedals he would have, by virtue of his own investigative urges, placed his free feet from time to time on the pedals and by those actions discovered earlier that it was leg-pushing on them that brought about movements of the vehicle”.

CONCLUSIONS AND FUTURE DIRECTIONS

A general conclusion that can be drawn from this thesis, is that studying muscle recruitment strategies of spontaneous motor actions (such as reaching movements) in infants, leads to valuable insights but also to additional questions. The postural challenge of reaching movements is much smaller than that of sudden balance perturbations, and leads to different responses: direction-specific adjustments are not consistently used during infancy, although their frequency does increase with age in typically developing infants. Variation is abundant; consequently, it is still unclear to what extent fine-tuning of the postural response (for example in recruitment order or anticipatory activation) is already adaptive. It would be interesting to do similar studies with spontaneous, but larger balance disturbances to answer this question. Such studies might also test our hypothesis that the infrequent use of direction-specific adjustments is related to the relatively minor intensity of the balance disturbance that is caused by the reaching movement. Perhaps infants at risk of cerebral palsy are also able to produce more direction-specific adjustments at this age during larger balance disturbances. In addition, variations in posture and position at the start of the reaching movement often made interpretation of the data difficult; combining EMG measurements with kinematic
and force plate data would facilitate investigating the relationship between muscle recruitment strategies and task constraints (such as the severity of the balance disturbance and the inherent variations in motor behaviour of young children). This would also make it easier to link successful muscle recruitment strategies to successful outcomes in terms of balance. Such information might then be used to help children with postural problems: for young infants by providing an environment that stimulates exploration of such strategies, and for older children by providing additional feedback on their postural strategies.
REFERENCES


