Measuring motor outcome in childhood

van Schie, P.E.M.

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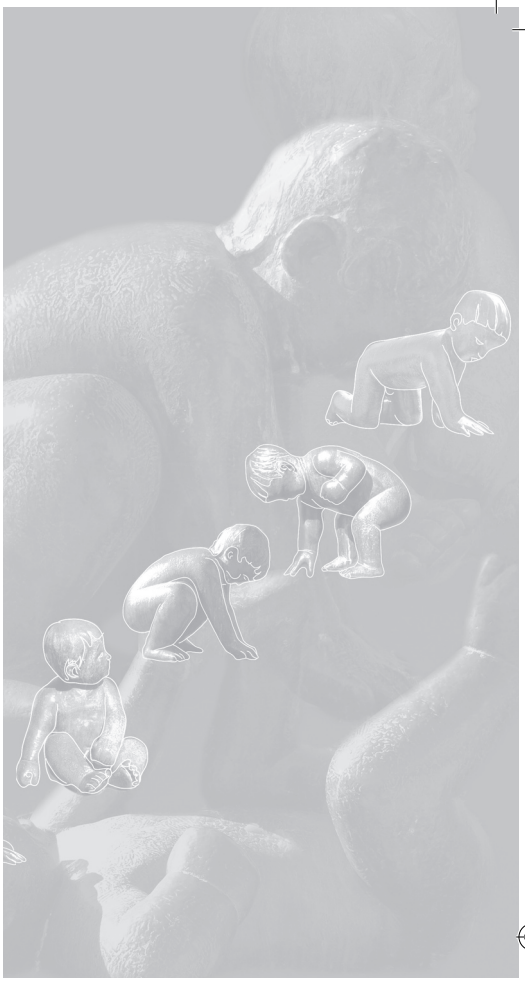

Summary

The long-term outcome of a child's development is always an intriguing issue, especially if the child was born after problems during pregnancy or delivery, or if the child has been treated with a relatively new intervention.

This thesis describes the results of studies focusing on the motor outcome of three different cohorts of children: 1) children, mostly small for their gestational age and born prematurely, from mothers with early-onset hypertensive disorders of pregnancy; 2) children born full-term with hypoxic-ischemic encephalopathy (HIE) due to perinatal asphyxia; 3) and children with cerebral palsy (CP) who were treated with Selective Dorsal Rhizotomy (SDR).

The main aims of this thesis are to describe motor outcome, mainly at the level of activity, in young children who were at risk for developmental problems or who had received an intervention, and to examine the predictive value of the measurement instruments that are most frequently used in childhood to forecast future motor outcome.

Chapter 1, the Introduction, describes the aims and outline of the thesis. In this chapter the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY) is introduced, and 'normal' motor development and issues concerning motor assessments are discussed. It also provides background information on the disorders in the study population (HIE and CP) and the SDR treatment. This is followed by a brief outline of the motor tests that are commonly used in clinical paediatric physical therapy practice to measure motor outcome in children.

Chapter 2 describes a study of the motor outcome of 175 infants. The aim of the study was to describe General Movements (GMs) at term age and three months post term, and their association with neurological examination performed at the same ages. Furthermore, the predictive validity of GMs at three months for outcome at one year corrected age was evaluated. Most infants were small for their gestational age and born prematurely from women with early-onset hypertensive disorders of pregnancy. This prospective study was part of a randomised controlled trial of pre-birth management strategies, the PETRA study (Pre-eclampsia and Eclampsia TRial Amsterdam), executed in a collaboration between the Departments of Obstetrics and Gynaecology of the Amsterdam Medical Center and the VU University Medical Center. We examined the prevalence of normal, mildly abnormal and definitely abnormal General Movements (GMs) and the results of neurological examination at term and three months corrected age in these infants. We investigated the association of GMs with neurodevelopmental outcome at one year, measured with the motor and mental scales of the Bayley Scales of Infant Development second version (BSID-II). We found an association between GMs and neurological examination results at term age, but at three months corrected age, GMs and neurological examination results were not related. GMs at three months were also not associated with abnormal neurological examination at one year, but they were associated with delayed motor outcome at one year. Neurological examination at three months was related to motor outcome at one year, but there was no association with neurological examination or mental outcome at one year. We concluded that, although reports in the literature suggest that the assessment of GMs at three months is useful for the prediction of adverse neurological outcome in high risk infants, we were not able to confirm this in our study. The assessment of GMs can be useful in the examination of high risk children, but should preferably be combined with other assessments such as neurological examinations.

In **Chapter 3** we investigated motor outcome at the age of one year in a cohort of 32 surviving children who were born full-term with perinatal HIE. Motor outcome was measured according to the Alberta Infant Motor Scale (AIMS), the motor scale of the BSID-II and a neurological examination (Neurological Optimality Score; NOS). We studied the relationships between, on the one hand, motor tests (AIMS and BSID-II motor scale) and neurological examination at one year, and on the other hand, the outcome of these tests and neonatal brain Magnetic Resonance Imaging (MRI).

We found that the AIMS and the BSID-II motor scale agreed in the classification of normal, mildly delayed or significantly delayed motor outcome at one year in approximately 75% of the children. All children with a normal motor outcome had a (near) optimal NOS, but not all children with a high NOS had a normal motor outcome. Children with a normal neonatal MRI had a normal or mildly delayed motor outcome at one year, but in children with abnormalities on the MRI motor outcome was variable at one year. We concluded that the two motor tests agreed on classification of motor outcome in the majority of cases, but not in all cases. Normal brain MRI is a good predictor for a normal or mildly delayed motor outcome, but abnormal MRI did not predict motor outcome at the age of one year. In line with these findings we suggest that a combination of motor tests and neurological examination at the age of one year should be used to assess outcome after HIE.

In **Chapter 4** we investigated motor and mental outcome at two years in our cohort of 32 children with HIE. We assessed the additional predictive value of the different tests (AIMS, BSID-II and NOS) at one year, in addition to prediction based on neonatal Sarnat staging and MRI, for both motor and mental outcome (measured with the BSID-II) at two years. We found that all children with Sarnat grade I had a good motor outcome, but 14% had a poor mental outcome. All children with normal MRI had a normal motor outcome, but 9% had a poor mental outcome. In children with Sarnat II or with neonatal MRI abnormalities the outcome was variable: approximately 50% had a normal motor outcome. Sarnat I and normal neonatal MRI were good predictors of a normal or only mildly delayed motor outcome. For children with Sarnat grade II and abnormal MRI, additional motor testing at one year improved the accuracy of predicting a poor motor and mental outcome at two years. In line with this finding, our conclusion was that motor testing at one year is helpful for the prediction of a poor motor outcome at two years. Nevertheless, a longer follow-up of these children is needed to establish how they will develop in the long term, acknowledging that a few studies suggest that even children with a good outcome at two years may have minor neurological dysfunctions or cognitive problems in later life.

In **Chapter 5** we evaluated the effect of SDR on gross motor function, self-care and gait pattern in a well-defined group of nine ambulatory children with spastic diplegia. These children were the first to undergo this operation in the Netherlands and were monitored extensively during the first year after SDR. Outcome was measured in the different levels of the ICF-CY. Gross motor function was measured with the Gross Motor Function Measure (GMFM-88) every two months, from 4 months before until one year after SDR. Self-care was assessed with the Pediatric Evaluation of Disability Inventory (PEDI) and gait pattern was assessed with the Edinburgh Visual Gait Score (EGS) at baseline and one year after SDR.

We found a significant improvement in the mean total GMFM-88 score between baseline and one year after SDR, indicating an improvement in gross motor function. These results were comparable to the effects reported in the literature. Using a single-subject research design, with repeated measurements over time, visual plots of individual cases showed significant change in GMFM-88 scores after SDR, compared to baseline measurements. These findings were confirmed by within-subject analysis with C-statistics. Functional skills and care-giver assistance for self-care, measured with the PEDI, also showed significant improvement in the entire study population. Improvement in gait pattern was found, in particular with respect to initial contact and heel-lift, resulting in a more normal EGS. We concluded that in this well-defined group of ambulatory children, SDR had a small but significantly positive effect on gross motor function, self-care and gait pattern.

In **Chapter 6** we evaluated the short-term (1 year) and long-term (3–8 years) effects of SDR in all 33 ambulatory children with spastic diplegia who had undergone SDR in the Netherlands. Gross motor function was assessed according to the Gross Motor Function Classification System (GMFCS) and GMFM-66. Spasticity was measured according to a modified Tardieu scale. Information about additional treatment and the opinion of the parents with regard to improvement in their child's functioning was obtained from questionnaires. We found that in both the short term (one year) and the long term (mean 6 years) after SDR, the mean GMFM-66 score had improved significantly from baseline, indicating improved gross motor function. However, most children had a stable short-term and long-term GMFCS level. In the short term, the children in GMFCS levels I–II improved significantly more on the GMFM-66 than the children in level III. In the long term there was no significant difference in mean GMFM-66 improvement between the children in GMFCS level I–II and III. Reference percentile data were used for comparison with the 'natural course' of gross motor development in children with CP. It has been stated that the chance of a change of more than 20 percentile data in one year in a cohort of children with CP is 20%. When comparing the individual percentile rankings, 29% of the children improved more than 20 percentiles in their GMFCS level between baseline and the one year follow-up, and 43% had improved more than 20 percentiles at the long term. We considered the effect of SDR to be an improvement on the expected natural course of gross motor development in a subgroup of children with CP, especially in the long term.

Spasticity had disappeared in the majority, although not in all children, and no relapse of spasticity was noted at the long-term follow-up. Almost one third of the children, mostly children in GMFCS level III, needed orthopedic surgery, especially operations to stabilise the feet. Approximately 40% of the children were treated with botulinum toxin-A injections after SDR, and most of these children were in GMFCS level III.

We concluded that SDR resulted in small short-term improvements, in particular for children in GMFCS levels I–II. In the long term, approximately 40% of the children showed improvements in their gross motor function, which was considered to be more than what could be expected, according the percentile ranking of their individual motor growth curve. Approximately 50% of the children needed additional surgery or botulinum toxin-A treatment. This also suggests that final conclusions about the effectiveness of SDR can only be drawn after the child reaches adulthood.

In **Chapter 7** the main findings described in the thesis were discussed, suggestions were made for future research, and clinical implications were formulated. Recommendations for future research include obtaining more knowledge about motor assessments, especially about the prognostic value of tests, and the development of adequate norms. A longer follow-up of children who are at risk for developmental problems is necessary, not only for research purposes, but also for the provision of care. Moreover, a longer follow-up of children is needed after SDR, acknowledging that the long-term effects of SDR are largely unknown. Cohort studies with a long-term follow-up should also focus on identifying the characteristics of children who benefit most from SDR.

Recommendations for clinical implications included longer follow-up for children who are at risk for developmental problems and children who are treated with SDR, which a core-set of measurement instruments, about which there is consensus in the Netherlands.

We recommend that all clinicians working with children with CP in multidisciplinary teams use the GMFM-66 in the assessment of children with CP, and should have the necessary knowledge about using the GMFCS and reference percentiles. Paediatric physical therapists can play an important role in (joint) diagnostics and the evaluation of interventions in multidisciplinary rehabilitation teams.