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Infants at very high risk of cerebral palsy

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SUMMARY

This thesis aims to unravel a part of the complex and intriguing puzzle of early brain development. Primary aim is to study effects of early intervention in infants at very high risk (VHR) of cerebral palsy (CP). CP is the most common cause of physical disability in pediatrics, originating from disturbances in early brain development. CP manifests with limited mobility due to difficulties with movement and postural control, often accompanied by other developmental disabilities, such as cognitive or behavioural problems. Secondary aims are a) increasing knowledge about VHR-infants' early development and factors influencing development, b) providing insight in contents of early intervention and associations between interventional elements and outcome, and c) evaluating use of motor function measures for VHR-infants.

PART 1: Factors that may affect outcome in very high risk infants

Chapter 2 reviews the literature on motor and cognitive outcome of VHR-infants with severe brain lesions. The studies included showed that severe brain injury is strongly associated with development of CP, most often in infants with cystic periventricular leukomalacia (86%) and less often in infants with term stroke (30%). In case of development of CP, usually unilateral lesions result in unilateral CP and bilateral lesions in bilateral CP. However, unilateral lesions may result in bilateral CP and vice versa. Intellectual disability has been described in 27-50% of the infants with severe brain lesions. Information about other influencing factors was little; sex specific outcome was only provided in few studies and socioeconomic class influences have been provided only sporadic.

Chapter 3 investigates contents of conventional infant physiotherapy during the last decades, as provided in the Netherlands. Within infants physiotherapy, developments occurred over time: over the years families are more involved, both in communication and in being educated by the physiotherapist. Involvement is most often in the form of training, providing feedback, exchange of information, or instruction. Neuromotor actions, such as facilitation, sensory experience and challenging the infant to self produced motor behaviour, did not change significantly over time. The results indicated that theoretical concepts have been implemented partly in practice.

PART 2: Early intervention in very high risk infants

In Chapter 4, results of the Vroegtijdig Interventie (Early Intervention) Project (VIP) are presented. In the VIP-study, infants at very high risk of CP, based on presenting with definitely abnormal general movements, were included. Infant motor outcome, measured by the Infant Motor Profile, was compared for the newly developed COPCA-intervention (Coping with and Caring for infants with special needs – a family centred programme) with

the control group TIP (Typical Infant Physiotherapy). Infants were randomly allocated to receive either COPCA or TIP between 3 and 6 months corrected age. In the Randomized Controlled Trial (RCT), infants receiving COPCA or TIP had similar outcome. After analysing contents of intervention, associations were present between interventional elements and outcome: some COPCA-related items were positively associated with outcome and some TIP-elements were negatively associated with outcome, especially in those infants who developed CP. However, only about a quarter of the included infants developed CP and the intervention period was only three months.

Therefore, a design for a new intervention study was developed, the LEARN2MOVE0-2 years (L2M0-2) study (chapter 5). Main differences compared with the VIP-study are: infants at higher risk for CP were included, i.e., mainly included on the basis of severe brain lesions, and the intervention period was longer: one year instead of three months. Besides infant outcome, also family outcome was included.

Results of the L2M0-2-study are presented in Chapter 6. In the L2M0-2-study more than half of the included infants developed CP. Comparable with the VIP-study, no differences at RCT-level for COPCA and TIP were shown. In contrast to the VIP-study, no associations between interventional elements and infant outcome were found in the L2M0-2-study. However, family outcome did show an association with intervention: family empowerment was positively associated with COPCA-related interventional elements.

PART 3: Measuring gross motor function in young infants with or at high risk of cerebral palsy

No 'gold standard' for measuring motor function in infancy is available. Often used measures in children with CP are the Gross Motor Function Measure (GMFM) and the Gross Motor Function Classification Measure (GMFCS). Both are known to be reliable and valid instruments, but generally applied in children who are already diagnosed with CP, i.e., usually after the age of 18-24 months. In Part 3 of this thesis, results and suggestions for use of the GMFM and GMFCS in infancy are provided.

Chapter 7 describes difficulties encountered with using the GMFM in infancy based on the first infants included in the L2M0-2-study. Suggestions for adaptations for use of the GMFM in infancy were provided, amongst others with eliminating or adapting items which are difficult to elicit in infancy and those items which require the ability to follow instructions. First results of the use of the adapted GMFM in the pilot study are promising and may better reflect the infant's actual motor function. However, further research is needed to define reliability, validity en responsivity of the suggested adaptations for the GMFM in infancy.

In Chapter 8, application of the GMFCS in infancy is discussed. Over the years, use of assisted or powered mobility is provided more and more at younger ages, also below the age of two years. The current GMFCS applies also to children below the age of two years,

but assisted mobility is not included in the description. Therefore, suggestions were made to implement assisted mobility also in the definition of the GMFCS for 0-2 years.

In conclusion, this thesis shows that infants with severe brain lesions are at high risk of developing neurodevelopmental disabilities, more than infants with only clinical signs of dysfunction. Contents of early intervention changed over the years. In the beginning of the 21st century, the family centred COPCA-intervention was developed, providing early intervention in infants with special needs. In the two studied intervention trials, infants who received COPCA or TIP have comparable outcome at RCT-level. However, specific elements of interventions were associated with outcome: in the VIP-project with infant outcome and in the L2M0-2-study with family outcome. Measuring outcome in high risk infants at young age is challenging, but a prerequisite for adequately measuring effects of early intervention. Therefore, some suggestions have been provided for adaptations in infancy for two often used measures: the GMFM and the GMFCS.

For future research, advancements in early detection and predicting outcome of infants at high risk may assist improved understanding of efficacy of provided interventions. Knowledge about contents of intervention is needed to study underlying working mechanisms. Being able to study outcome of early intervention and reflecting infants' actual functioning, measures in infancy need further investigation and optimization. Studies should not only focus on infant outcome, but also on family outcome, which may be more malleable than the infant brain when the infant has a severe brain injury.

