Early intervention in infants at high risk for developmental motor disorders

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Early intervention in infants at high risk for developmental motor disorders

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Contents

CHAPTER 1

INTRODUCTION

In approximately 8% of all live births in the Netherlands preterm delivery occurs (CBS 2008). Advanced technology in neonatal intensive care over the past 20 years has increased the survival of preterm and high-risk full term infants (Hack and Fanaroff 2000, Stoelhorst et al 2005). Follow-up studies, however, have shown an increased incidence of motor disability later in life, ranging from developmental coordination disorder to cerebral palsy (CP) (Hack and Fanaroff 2000, Marlow 2004, Stoelhorst et al 2005, Fawke 2007). To optimize developmental outcome of these infants extensive early intervention programmes have been developed over the years. In this thesis I will focus specifically on the effects of paediatric physical therapy programmes that have been applied to infants up to 18 months corrected age who are at risk for a developmental motor disorder.

The aim of this thesis is to contribute to the body of knowledge about the effects of early physiotherapeutic intervention for young infants who are considered to be at risk for a developmental motor disorder. Existing programmes are reviewed and a new physiotherapeutic intervention programme called 'COPCA' (COPing with and CAring for infants with special needs - a family centred programme; Dirks and Hadders-Algra 2003) is evaluated. The goal of this evaluation is twofold. First, the contents of COPCA are analysed and compared to the existing physiotherapeutic approach. And second, the effects of the COPCA intervention on developmental outcome are evaluated.

In this introductory chapter brief background information will be provided about the key subjects in this thesis. In the following paragraphs information about early intervention, developmental motor disorders of the infants in the studied cohort, and the main influences on paediatric physical therapy in the Netherlands will be provided. The chapter concludes with an outline of the individual studies of this thesis.

Early Intervention

Young infants who are discovered to be at risk for developmental motor disorders are often referred to early intervention services. Early intervention (EI) consists of multidisciplinary services provided to children from birth to 5 years of age (Shonkoff and Meisels 2000). The aims pursued with EI are remedial or preventative in nature and comprise the following: 1) promote child health and well-being, 2) enhance emerging competencies, 3) minimize developmental delays, 4) remediate existing or emerging disabilities, 5) prevent functional deterioration and, 6) promote adaptive parenting and overall family functioning (Shonkoff and Meisels 2000). These goals are accomplished by providing individualized developmental, educational and therapeutic services for children in conjunction with mutually planned support for their families. EI can be provided in the hospital, in a rehabilitation centre, in the physical therapy practice, at home or in a combination of these four. Although EI may start at any moment between birth and 5 years of age it is advocated to intervene as early as possible. It is recognized that only through early identification and appropriate programming infants can reach their full potential. In a recent review on brain development de Graaf-Peters and Hadders-Algra (2006) recommended that, based on developmental processes, the best time window for EI in terms of active stimulation of the infant lies between 40-44 weeks post menstrual age and 15 months postnatally. In this period, processes of dendritic outgrowth and synapse formation are highly active which offer the best opportunities to reconnect and find functional solutions than later periods (Kolb et al 2001, de Graaf-Peters and Hadders-Algra 2006). Active stimulation consists of activities which induce active responses of the infant, for example offering toys to stimulate head balancing exercises in prone position. De Graaf-Peters and Hadders-Algra (2006) suggest also that EI prior to 40-44 weeks post menstrual age should be restricted to forms which aim at mimicking the intrauterine environment in order to reduce stress. Other advantages of intervening early are possible beneficial effects on the family. From the literature it is known that families who encounter problems with their infant's development experience higher levels of stress (Singer et al 1999, King et al 2004, Muller-Nix et al 2004, Butcher et al 2008). EI can help to minimize the impact of developmental problems on the family.

Summarizing, the research field of EI is very broad. It covers a wide age span and a whole range of services. Therefore in this thesis the term early intervention is limited to paediatric physical therapy for infants up to 18 months corrected age who are at risk for a developmental motor disorder.

Developmental motor disorders

Cerebral palsy and developmental coordination disorder are two important forms of developmental motor disorders. Cerebral Palsy (CP) is defined as "a group of disorders of the development of movement and posture causing activity limitation, which are attributed to non-progressive disturbances that occurred in the developing foetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, behaviour, by epilepsy and by secondary musculoskeletal problems" (Bax et al 2005, Rosenbaum et al 2007). The prevalence of CP in the Netherlands is around 1 or 2 children per 1000 live births with an incidence which is higher in males than in females (1.33:1) (Wichers et al 2005). CP is the result of an injury or abnormal development of the brain which may occur during pregnancy, birth or within the first two to three years of life. In many cases, the exact cause of CP is unknown (Rosenbaum 2003). Currently, problems occurring during intrauterine development, congenital disorders, hypoxic-ischemic insults, and preterm birth are considered to account for the majority of the cases with CP (Moster et al 2001, Jones et al 2007). Infants born prematurely have an increased risk for intraventricular hemorrhage (IVH) or periventricular leukomalacia (PVL) resulting in injury to the periventricular white matter of the brain. PVL is a good predictor for CP, with CP developing in 80% to 85% of the infants (Perlman et al 1996, Jones et al 2007). All children with CP suffer from some degree of motor impairment which in most cases manifests itself before 18 months of age and persists throughout the lifespan. During early infancy CP cannot be diagnosed, because in many cases the characteristic features belonging to CP have not manifested themselves. The way CP affects each individual will vary widely. Characteristics may include a lack of coordination, spasticity, involuntary movements, muscle weakness, increased tendon reflexes, different walking patterns (i.e. scissors gait, toe walking, wide-based gait), dysarthria, difficulty with

gross and fine motor skills, and abnormal perception and sensation.

The clinical picture of CP evolves with time, development, learning, training, therapies, and other factors (Bower and McLellan 1994, Bax et al 2005). Therapy therefore focuses on helping the individual to maximize his or her potential (Scrutton 1984). In general it is believed that the earlier treatment starts the better chance children have of learning new ways to accomplish challenging daily tasks. While this seems intuitively obvious, strong scientific support is lacking to support this assumption (Damiano 2006).

Developmental coordination disorder (DCD) denotes children whose performance in daily activities that require motor coordination is substantially below what can be expected given the child's chronological age and intellectual ability, in the absence of other neurological disorders (APA; DSM-IV; 2000). The disturbance in the performance of daily activities interferes with academic achievement or activities of daily living. Estimates of the prevalence of DCD range from 5% to 15% in the primary school population (Wilson 2005). Most studies on children with DCD report a higher prevalence in boys than girls, but the actual sex ratio is variable and depends on the method used to identify children (Missiuna et al 2006).

Children with DCD not only differ in the motor skills they experience problems with, or the underlying processes that are causing the deficit, but also in the extent to which they suffer from co-morbidities such as attention, behavioural or emotional problems (Dewey et al 2002, Gillberg and Kadesjö 2003). Studies on the underlying processes have shown that children with DCD can have deficits in many areas of performance, including motor planning, motor programming, postural control, proprioceptive or kinesthetic deficits, and visual spatial processing (Niemeijer 2007). About half of children with DCD will not outgrow their motor problems (Losse et al 1991, Cantell et al 2003).

DCD is difficult to diagnose before school-age. Clumsy motor behaviour becomes more evident with increasing difficulty of motor tasks. Research has shown that a relation exists between the presence of neurological dysfunction in infancy and DCD (Jongmans et al 1997, Hadders-Algra 2003). A few studies have shown that early identification (i.e. before school-age), effective intervention and vocational counselling are important for children diagnosed with DCD to diminish the negative experiences that can affect their later academic and social life (Cantell et al 2003, Missiuna et al 2003).

Advances in paediatric physical therapy

In the early 1900s European physical therapists were trained in manipulation by a London internist and orthopaedic surgeon Dr. James Cyriax. CP at this time was considered to be a disease which they tried to cure with orthopaedic surgery. Over the years many different intervention programmes have been developed for young children with CP varying from different types of physical therapy and occupational therapy to the use of drug such as baclofen or botox, orthotic devices, etcetera. The variety in approaches might result from the diversity in effect that CP has on the daily functioning of the child (Mayston 2004). In this introductory chapter a brief overview of the main influences on paediatric physical therapy for infants with or at risk for CP is provided. In the Netherlands the main influences come from Neurodevelopmental Treatment (NDT), Vojta, Sensory Integration Therapy, and more recently from functional therapy. The intervention programmes differ in their treatment methods, but all aim at 'leading the children with cerebral palsy … towards the greatest degree of independence possible, and so to prepare them for as normal as an adolescent and adult life as can be achieved. This is the aim of all schools of treatment' (Bobath and Bobath 1984, p.6).

NeuroDevelopmental Treatment

The NDT approach was developed by Berta and Karl Bobath in the 1940s. The basic assumption of NDT was that sensorimotor problems in CP arise from central nervous system dysfunction, which interferes with the establishment of normal motor development. Inhibition of spasticity and facilitation of normal posture and motor behaviour were the main aims of treatment, which the Bobaths tried to accomplish by tone influencing patterns with the use of various therapeutic handling techniques (Bobath and Bobath 1984, Mayston 2001). The normal developmental sequence was advocated as a framework for treatment (Bly 1991, Howle 2002). In the sixties Elizabeth Köng and Mary Quinton started to adapt and develop the techniques for early treatment of babies based on the Bobath concept (Köng 1966, Quinton and Nelson 2002).

During the years the NDT approach evolved from a practice-based concept to a more theory-based and partly evidence-based approach (Bly 1991, Palmer 1997, Mayston 2000, Howle 2002). This means that based on new theoretical constructs and practical knowledge NDT gradually evolved and caused a shift in goals, treatment techniques and treatment application (Howle 2002). The original goal 'inhibition of spasticity and facilitation of normal posture and motor behaviour' to give the child the opportunity to move more normally nowadays has shifted to 'optimize function' (Howle 2002). This implies that NDT intervention aims to prepare and guide the children towards their greatest possible independence in everyday functioning (Bobath and Bobath 1984).

Based on experience and new insights in neurosciences the emphasis on certain treatment techniques changed. In the early years specific postures were imposed on the child with the aim of achieving a normalization of muscle tone. The 'reflex-inhibiting' postures and tone influencing patterns indeed did reduce spasticity during treatment, but lacked carry-over effects into movement and function (Mayston 2001, Howle 2002). Next, the Bobaths promoted the use of 'key points of control', i.e. special combinations of hand placement on the body that allowed the therapist to control the child's sensorimotor input and motor output. These handling techniques had the dual aim of inhibiting abnormal movement patterns and facilitation of normal movements (Bobath and Bobath 1984, Butler and Darrah 2001, Howle 2002). Moreover, therapeutic handling from key points of control made it possible for the child to - systematically and increasingly - take over control of his/her own movement.

Over the years therapists gradually became more aware that their treatment had not automat-

ically carried over into activities of daily life. As a consequence, preparations for functional tasks currently are carried out in settings where children live, play and learn (Bobath and Bobath 1984, Butler and Darrah 2001, Howle 2002). The principles for the NDT baby treatment (Bly 1999) aim at helping the infant in its development by the use of sensorimotor experiences (handling) and instructing the caregivers. In this regard caregivers play a specific role in NDT treatment. They have a role as co-therapist (Finnie 1996). Through guiding and training of caregivers, the therapist tries to establish carry-over from treatment into activities of daily life (Bobath and Bobath 1984). The hands of the therapist facilitate, assist or control the infants' posture and movements until the infant can take over. The treatment consists of a sequence of developing, kinesiological, sensory and environmental factors.

Treatment techniques

Tone influencing patterns are the primary intervention strategy of NDT that are achieved through various therapeutic handling techniques. The use of a tone influencing pattern inhibits abnormal activity, while at the same time the way is paved for more normal movements (e.g. facilitation) (Howle 2002). However, in 1965 the Bobaths already acknowledged that: "unless you stimulate or activate your patient in a way in which new activities are possible, you have done nothing at all. So the tone influencing patterns as such are only the very first step in treatment, though they are very important" (Mayston 2005). Thus, during NDT treatment inhibition and facilitation always act as interactive partners in order to prepare the body for the performance of functional tasks. Furthermore, therapeutic handling techniques allow the therapist to provide sensorimotor experience, to feel responses to changes in posture and movement, to gain control of muscle tone, to influence the alignment of the body, to direct the attention of the child and to recognize the moment when the child can become independent of assistance (Howle 2002). Therapeutic handling consists of the following interactive components:

1) Key points of control are predefined parts of the body on which the hands of the therapist are placed to influence directly the area beneath the hands of the therapist and other parts of the body. The key points of control allow the therapist to inhibit abnormal movement patterns and to facilitate normal movements simultaneously (Bobath and Bobath 1984). Key points are located all over the body and are used interchangeably and in combinations. During treatment there is a shift in the use of key points from proximal (e.g. shoulder, pelvic girdle) to distal (e.g. extremities) to give the child the opportunity to gradually gain control over his movements.
2) Facilitation is the strategy of therapeutic handling that makes a posture or movement more likely to occur. It can modify postural control by increasing degrees of freedom, supporting a body segment, or activating the postural system. Placing the child in an imposed posture creates a possibility to promote the occurrence of normal movement patterns (Howle 2002) (p.255-256).
3) Inhibition originally referred to reducing tone and abnormal reflex activity resulting from dysfunction in the central nervous system. Currently, inhibition refers to the reduction of specific underlying impairments that interfere with function (Howle 2002) (p.257). Specific goals are to gain control of muscle tone, to decrease excessive co-activation patterns and to prevent the occurrence of atypical postures.

As NDT is the most widely applied method in the treatment of infants with or at risk for developmental disorders, many studies are undertaken to demonstrate its effectiveness. In three recent reviews (Butler and Darrah 2001, Blauw-Hospers and Hadders-Algra 2005, Spittle et al 2007) attention was paid to the effectiveness of NDT. All three concluded that there was no advantage to NDT over other alternatives to which it was compared. No clear evidence was found that NDT was beneficial for motor development or functional motor activities. Also other potential benefits such as the enhancement of cognitive development or improved caregiver-infant interaction could not be demonstrated. Possible explanations that are provided for the scarceness of evidence are for instance the ongoing change of the constructs of NDT over time, the skill level of the therapist and the impossibility to deliver treatment in a standardized manner (Butler and Darrah 2001). While for research purposes the treatment setting can be standardized, the child with CP and his family can never be. Moreover, CP is a highly heterogeneous condition which is complicated by ongoing change in the children due to processes of growth and maturation. The change of the NDT constructs over time also includes that techniques of NDT were more often used together with techniques from other approaches that gained influence in the treatment of children with developmental disorders, like for example the Vojta concept and sensory integration therapy.

Treatment according to Vojta

The Vojta concept, also called reflex locomotion, was developed during the 1950s by Vaclav Vojta based on empirical observations. It was implemented in the Netherlands in the 1970s and is still applied, especially in the southern part of the country. The method is primarily considered to be a facilitatory approach that can be applied to all neuromuscular conditions, not just CP. Goals of reflex locomotion are: 1) to facilitate the automatic control of the body position, 2) to facilitate the uprighting mechanism of the body, and 3) to stimulate coordinated muscle activity. These skills are disturbed in children with brain lesions or developmental motor disorders.

The concept is based on the observation that through specific peripheral stimulation normal motor reactions throughout the body can be elicited when the patient is placed in a specific position. Vojta believed that the persistence of newborn reflexes in a child with CP interferes with normal postural development. It is assumed that the activation of newborn reflexes through stimulation of the global reflex pattern facilitates the development of reflex locomotion (Vojta 1984, Stanger and Oresic 2003, Mayston 2004).

Treatment techniques

The Vojta concept actually consists of two parts: clinical evaluation and treatment. Before the physiotherapist can start with the reflex locomotion treatment, he can use the Vojta concept as

a clinical evaluation to define the goals for the treatment. The clinical evaluation is divided into three parts. The first part is the study of the automatic postural reactivity. To discover potential problems in the organization of automatic responses by the CNS, reactions to sudden postural changes of the body, i.e. disturbances of the equilibrium position, are tested. This helps to identify the degree of development at time of the examination. The second part consists of the kinesiologic analysis of spontaneous motor function. The placement of the extremities at specific angles is important to maintain the equilibrium of the body. The third part is reflexology. A series of reflexes completes the examination to refine the immediate evaluation but also to specify the prognosis (Vojta 1984).

In the practical application reflex locomotion utilises two so-called coordination complexes: the reflex creeping and rolling. Vojta identified a number of reflex points which can be used to stimulate the human body to crawl and turn. From several start positions (supine, prone or side-lying) the flow of motor responses is provoked by giving pressure on specific body parts, the so-called 'zones'. During the reflex locomotion it is essential that the placement of the extremities at specific angels is optimal. The motor responses that arise during the application of the reflex locomotion are called 'global patterns'. Global implies that skeletal muscles throughout the body are activated in a coordinated manner and that the central nervous system (CNS) is addressed at all it's regulatory levels (Vojta 1976).

Caregivers play a specific role in treatment according to Vojta. They are thoroughly instructed to carry out the patterns of reflex locomotion. Optimally, the therapy should be carried out 4 times a day. The child is not actively involved in the treatment.

Vojta states that the best results of reflexlocomotion are achieved in babyhood when the plasticity of the brain is greatest. He claimed that pathological motor patterns are already present but not yet fixated and that the opportunity to gain access to normal movement patterns is merely blocked (Vojta 1976, 1984).

The effect of treatment according to Vojta is evaluated in only a few studies. Blauw-Hospers and Hadders-Algra (2005; see Chapter 3) showed that there was no evidence that treatment according to Vojta supports the development of infants at high risk for developmental motor disorders. The Vojta approach has been criticized for the emotional stress it may produce in child and family (d'Avignon et al 1981).

Sensory Integration Therapy

Sensory integration therapy was developed during the 1960s by Jean Ayres. She was interested in the way in which sensory processing and motor planning disorders interfere with activities of daily living and learning. Sensory integration is the process by which people register, modulate, and discriminate sensations received through the sensory systems to produce purposeful, adaptive behaviours in response to the environment (Ayres 2005). Sensory integration therapy is based on the assumption that learning is dependent on the ability to take in sensory information from

the environment, to process and integrate the sensory inputs within the central nervous system and to use this information for the planning and production of organized behaviour (Ayres 1972, Mayston 2004). Sensory integration focuses on the five sensory areas: vestibular, tactile, proprioceptive, visual and auditory information. The goal of intervention is to improve the ability to process and integrate sensory information and to provide a basis for improved independence and participation in daily life activities, play, and school tasks (Schaaf and Miller 2005). Although originally sensory integration therapy was developed to treat children with learning disabilities, Ayres recognized the utility of the theory for other clinical populations (Schaaf and Miller 2005). As a significant amount of the children with CP also experience sensory impairments, sensory integration may assist the child during the intake and processing of sensory information and thus enhance function.

The theory of Sensory Integration is based on principles from developmental psychology, education, neuroscience, and occupational therapy. It entails four main principles that are: 1) sensorimotor development is an important substrate for learning; 2) the interaction of the individual with the environment shapes brain development; 3) the nervous system is capable of change (plasticity); and 4) meaningful sensory–motor activity is a powerful mediator of plasticity (Schaaf and Miller 2005).

Treatment techniques

The therapy is driven by four main principles: 1) Just right challenge (the child must be able to meet the challenges through playful activities), 2) Adaptive response (the child adapts behaviour to meet the challenges presented), 3) Active engagement (the child wants to participate because the activities are fun), 4) Child-directed (the child's preferred activities are used in the session). Therapy provides opportunities for the child to engage in sensory motor activities rich in tactile, vestibular, and proprioceptive sensations. A key point for therapy success is the inner drive of the child (Ayres 2005). Based on the tasks the child would like to master, the therapist designs the environment which has to be appealing to the child and which is adapted to the child's developmental level. The treatment setting looks like a play-area with equipment designed to provide the required form of stimulation. During these play activities the child can actively explore its environment. The therapist assists the child to accomplish the task by manipulating the environment in such a way that the most adaptive response is elicited. Fun and success are prerequisites for feelings of competency that are needed to achieve progress in the processing of sensory input. During the activity the therapist observes the child's responses and increases or decreases the sensory and motor demands when necessary.

Over the past 30 years a number of studies have been conducted to investigate whether sensory integration therapy is effective. The majority of studies have focused on the use of sensory integration therapy with children with learning disabilities and have aimed at improving motor skills, academic performance, behavioural performance and/or sensory and perceptual skills. Two re-

views (Vargas and Camilli 1999, Schaaf and Miller 2005) reported that the evidence for the effects of sensory integration is inconclusive. Bumin and Kayihan 2001 conducted a study to examine the effects of sensory integration therapy in children with spastic diplegia. They found that children who received the sensory integration programme performed better compared to a no treatment control group. No studies were found which evaluated the effect of sensory integration therapy for infants.

Functional Therapy

In the past decades the assumptions of the traditional treatment approaches have been questioned in a considerable number of studies. Evidence for the effectiveness of the approaches is inconclusive. A number of studies have shown that treatment often was not automatically transferred into meaningful activities of daily life (Bobath and Bobath 1984, Butler and Darrah 2001, Ketelaar et al 2001). This finding initiated a conceptual shift in treatment from focusing on normality to functionality. Nowadays, the concept of functional therapy is increasingly emphasized in literature on interventions for children with CP.

The theoretical background of the functional approach is formed by the dynamic systems theory (Thelen and Smith 1994) and family-centred service. Functional therapy consists of an approach guided by principles of motor learning and function (Ketelaar et al 2001, Ekström-Ahl et al 2005). Exploration and selection in finding solutions for new task demands in combination with the adaptation to changes in the environmental context are considered to be critical parts of motor learning (Ketelaar et al 2001). Main features of a functional approach to therapy are: 1) Focus on the improvement of functional tasks which are problematic in daily life, 2) The child acts as an active participant of the therapy, who gets the opportunity to actively explore his possibilities and to find the best strategies and solutions for a functional task, 3) Training should take place in a context specific environment, 4) Focus on assisting the child and his family to achieve more satisfactory performance of daily activities (Harris 1990, Law et al 1998, Ketelaar et al 2001), 5) Caregivers are involved in all stages of the programme (from goal-setting and implementation in daily life to evaluation).

In the last decades the involvement of caregivers in the care process has become the basis of the care approach in rehabilitation. This concept of family centered care (FCC) is described as a holistic approach towards service delivery, in which the unique strengths, resources and set of needs of each child and its family form the base for a highly individualized and dynamic model of care (King et al 2004, MacKean et al 2005). Keywords in FCC are partnership, information exchange and respectful and supportive treatment (King et al 1997, Rosenbaum et al 1998). Caregivers are considered as equal partners who work together with the rehabilitation team to optimize their child's care process. This implies that caregivers are involved in all stages of the rehabilitation program, from goal-setting and implementation to evaluation. However, (Law et al 2003) state that true FCC gives caregivers the opportunity to decide their own level of involvement. The intervention programme must be adapted to families' capabilities, their situation and

daily schedule and not vice versa. This view is supported by Nijhuis and coworkers (2007) who showed that to provide true FCC a family-specific evaluation of FCC, i.e. making an accurate inventory of the specific needs and wishes of the individual families, is essential to avoid possible discrepancies between the views of caregivers and health care professionals.

Research has indicated that the involvement of caregivers in the care process can be associated with positive effects both on the development of the child (Ketelaar et al 1998, Law et al 1998, King et al 2004) as well as on the caregivers in terms of an increase in parental skills, improved well-being and a decreased level of parental stress (King et al 1996, Rosenbaum et al 1998, Jansen et al 2003, King et al 2004, Raina et al 2005). Caregivers can play a major role in assisting their children to reach their maximum potential by training the transfer of functional skills form one context to another. Moreover, the involvement of caregivers in their child's intervention may improve their understanding of their child's development (Palmer et al 1988). From the literature it is assumed that daily home-based physical therapy provided by the parents is more efficient than physical therapy given by a therapist once a week for a half hour (Ketelaar et al 1998).

Evidence for the efficacy of functional therapy is slowly increasing. One of the first studies that evaluated a somewhat functional approach was an often cited study by Palmer et al (1988). Forty-eight infants, aged 12 to 19 months, were randomly assigned to receive an infant stimulation programme including motor, sensory, language and cognitive activities of increasing complexity or NDT. The infant stimulation programme turned out to be more beneficial for developmental outcome than the NDT approach. Another study which showed that functional physical therapy was beneficial for children with cerebral palsy was written by Ketelaar et al (2001). They showed in 55 children with CP aged 2 to 7 years that the children who received the functional therapy programme (n=28) after the intervention period improved more on functional skills in daily situations than the control group. In 2005 a pilot study was published which evaluated the effects of a functional therapy programme on motor development and functional outcome measures of preschool children with CP (Ekström Ahl et al 2005). Fourteen children aged 18 months to 6 years participated in the five-month intervention. The study showed that after the programme the children had better gross motor function and performed better on daily activities. Also in relation to the children's mobility a decrease was found in the amount of caregiver assistance. A major drawback of this pilot study however was the lack of a control group. At the moment a large multi-centred RCT is running on the effects of a functional therapy programme for young children with CP in Canada (Law et al 2007). The study aims at an inclusion of 220 children with CP aged 12 months to 5 years.

Initially, the functional approach was developed for children with developmental disorders. Applying this concept to young infants (less than 18 months of age) implies that therapy mainly focuses on the encouragement of self produced motor behaviour (Resnick et al 1987, Palmer et al 1988, Ketelaar et al 2001, Ekström-Ahl et al 2005). Infants act as active participants of the therapy (Damiano 2006, Callahan et al 2006) who get the opportunity to actively explore their motor pos-

sibilities and to find an appropriate solution for any given motor task. The therapist coaches the caregiver to stimulate the infant to perform tasks that are both challenging and meaningful with appropriate success and failure rates (Dirks and Hadders-Algra 2003). The focus lies on assisting the family to achieve more satisfactory performance of daily activities (Harris 1990, Law et al 1998, Ketelaar et al 2001).

Concluding remarks

Paediatric physical therapy for infants at high risk for a developmental motor disorder can be considered as a field in development. Based on findings that evidence for the effectiveness of the traditional EI programmes (NDT, treatment according to Vojta, and Sensory Integration Therapy) is limited, shifts in the application of therapy were initiated. The most important advances are functionality instead of normality, the infant as an active participant of the therapy and the inclusion of the family.

Aims and outline of this thesis

The advances in paediatric physical therapy show that the physiotherapeutic guidance of infants with or at high risk for a developmental motor disorder is very diverse. The evidence for the effectiveness of the intervention programmes is inconclusive. Most studies have evaluated the effects of early intervention at the time during infancy or preschool age when the condition is already expressed in dysfunction. As mentioned before, the term early intervention in this thesis is limited to paediatric physical therapy for infants up to 18 months corrected age. Therefore, the first aim of this thesis was to review the effects of early physiotherapeutic intervention on motor and cognitive development of infants with a high risk for developmental motor disorders aged less than 18 months. Two systematic reviews were performed to gain more insight in the currently available evidence on early intervention. In these reviews specific attention is paid to unravelling elements in intervention programmes which might contribute to a beneficial effect on motor or cognitive development. The results of these reviews are presented in *Chapter 3 and 4*.

The results of both reviews indicated that intervention programmes with an onset after term age which focus on the stimulation of self-produced motor behaviour can exert a beneficial effect on motor and cognitive development. This knowledge served as a base for the development of a new type of physiotherapeutic intervention, COPCA. COPCA is the abbreviation of Coping with and Caring for infants with special needs – a family centered programme (Dirks and Hadders-Algra 2003). The rationale behind the COPCA programme is presented in *Chapters 2 and 6*. To evaluate the effects of COPCA on the development of infants at high risk for a developmental disorder an early intervention project VIP (in Dutch "vroegtijdig interventie project") was designed. In the VIP project the effects of the COPCA intervention on neuromotor and cognitive development were evaluated and compared to the effects of the traditional approach towards traditional infant therapy (TIP). For a description of the study design, subjects and outcome measures involved in the early intervention project VIP see *Chapter 2*.

The second aim of this thesis was to evaluate the effects of COPCA and TIP on developmental outcome. The results of this evaluation are presented in *Chapter 4, 7, and 8*. In *Chapter 4* some preliminary results on developmental outcome for the first half of the study group are described. *Chapter 7* has a twofold focus. In the first part of the study the effects of COPCA and TIP on the developmental outcome at 6 and 18 months are described. In the second part the findings of the randomized controlled trial were extended with the findings on the contents of the intervention (described in Chapter 5 and 6). In this way we attempted to identify whether changes in developmental outcome were associated with the contents of the intervention or that they could be explained by other factors. *Chapter 8* discusses the outcomes of the VIP project including some data which have not been published earlier.

The third aim of this thesis was to evaluate the application of COPCA in daily practice by means of video analysis. This process evaluation is a prerequisite for the implementation of COPCA within paediatric physiotherapy. Through detailed analysis of videos of physiotherapeutic intervention sessions we gathered knowledge on the actual contents of both the COPCA and TIP intervention. This analysis will help to discover the effective and ineffective elements in the interventions and will consecutively result in knowledge about the implementation of intervention by different paediatric physical therapists in different settings. *Chapter 5* introduces an observation protocol which we developed for the analysis of actions that physiotherapists use during treatment. Eight main categories of physiotherapeutic actions were defined to explore the contents of COPCA and TIP. *Chapter 6* reports the contents of TIP and COPCA. Here the similarities and differences between COPCA and TIP are lined out.

The thesis concludes with a general discussion of the reported findings focusing on implications for further research and practice.

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CHAPTER 2

THE 'VROEGTIJDIG INTERVENTIE PROJECT' (VIP)

Dirks and Hadders-Algra (2003) developed a new physiotherapeutic intervention programme, named COPCA, for infants at high risk for a developmental motor disorder. COPCA stands for COPing with and CAring for infants with special needs – a family centred programme. The COPCA intervention is based on new insights in the field of education and family care and in the normal and abnormal development of the central nervous system. The effects of COPCA on the development of infants at high risk for a developmental motor disorder were evaluated in the early intervention project VIP (in Dutch "vroegtijdig interventie project"). The VIP project was started in 2003 and the results are presented in this thesis (see *Chapter 4,5,6,7, and 8*). Infants who had been admitted to the Neonatal Intensive Care Unit (NICU) of the University Medical Center Groningen between March 2003 and May 2005 and showed definitely abnormal General Movements (GM) were eligible for this study. The presence of definitely abnormal GMs indicates a high risk for developmental motor disabilities, such as cerebral palsy (CP) (Prechtl 2001, Hadders-Algra 2004). Infants with severe congenital anomalies and infants whose parents had an inappropriate understanding of the Dutch language were excluded from the study. The VIP project was designed as a randomized controlled trial. Through block randomization infants were assigned to the experimental group who received the COPCA intervention or to the control group who got the traditional form of infant physical therapy (TIP). The second goal of the VIP project was to evaluate the application of COPCA in daily practice by means of video analysis. This process evaluation is a prerequisite for the implementation of COPCA within paediatric physiotherapy at a later stage.

In the next section a brief outline of the TIP and COPCA intervention is provided. The remaining sections of this chapter describe the study design, subjects and outcome measures involved in the early intervention project VIP. The outcomes of the VIP project are described in *Chapter 4, 5, 6, 7, and 8*.

TIP

TIP consists of infant physical therapy as it is nowadays applied in the Netherlands. TIP includes treatment strategies from a variety of physiotherapy programmes such as NDT, treatment according to Vojta and the Functional approach. For a description of these programmes see Chapter 1. For the most part TIP consists of the implementation of the 'living concept' of neurodevelopmental treatment (NDT; Howle 2002). Influences from the functional approach, such as the encouragement of self produced motor behaviour, are more and more incorporated into the treatment. Due to the different influences which are incorporated in TIP a large heterogeneity in treatment application can be expected.

COPCA

COPCA is a relationship family focused intervention programme built on two theoretical components. The first component is based on the transactional model of development (Sameroff and Chandler 1975, Fiese and Sameroff 1989) and recent findings in the field of education and family care (Dale 1996, Law et al 1998, Rosenbaum et al 1998). The second component, a neurodevelopmental one, is based on the principles of the Neuronal Group Selection Theory (NGST; Edelman 1993).

An important base of COPCA is formed by the transactional model of development (Sameroff and Chandler 1975, Fiese and Sameroff 1989) and new insights in the field of education and family care (Dale 1996, Law et al 1998, Rosenbaum et al 1998). In the transactional model development is seen as a result of a continuous dynamic interplay among child behaviour, caregiver responses to the child's behaviour, and environmental variables that may influence both the child and the caregiver. Therefore in COPCA optimizing the interaction between caregiver and child is a key factor of the intervention. The physiotherapist can be seen as an environmental factor that may influence both the child and the caregiver. The physiotherapist acts as a coach who respects the autonomy of the family and intends to make him- or herself superfluous. Caregivers are key-persons in the COPCA programme. The physiotherapist and the family are partners, in which the family defines the priorities for intervention, while the therapist coaches the family throughout the intervention period (Law et al 1998). The PT provides information on infant development, the specific problems of infants with neurological dysfunction, basic principles on infant education. Specific ways in which development may be promoted and problems may be tackled. By means of a continuous dialogue with the PT, caregivers develop their own ways to cope with the problems of the infant.

The second component of COPCA is based on the principles of the NGST. The NGST emphasizes that development is the consequence of a complex interaction between genetic information and environmental influences. According to the NGST development is characterized by two phases of variability, i.e. primary and secondary variability. During primary variability the child explores all variations of motor possibilities that are available in the nervous system. In this phase the child is not yet able to adjust his behaviour to external conditions. In typically developing infants this phase is characterized by abundant variation. At function specific ages the infant reaches the phase of secondary variability. The child gradually learns to select the most efficient solution for a given task out of his motor repertoire. This selection is based on trial and error experiences (Hadders-Algra 2000a). This means that during the phase of secondary variability the child learns to adapt his/her motor behaviour to specific situations. Infants with a pre- or perinatally acquired brain lesion resulting in a developmental motor disorder such as CP have a reduced repertoire of motor strategies available. This is already expressed during the first postnatal months in a limited repertoire of general movements and continues when goal-directed motility emerges (Hadders-Algra 2000b). In addition, these infants have problems with the selection of the most appropriate solution for a certain task out of the repertoire due to deficits in the processing of sensory information (Hadders-Algra 2000b). Therefore COPCA aims to promote variation in motor behaviour and trial-and-error experiences by the means of play. The ultimate goal is to provide an infant with a larger diversity in neuronal networks that will help to find an appropriate solution for any given motor task.

Subjects and Procedure

The study groups of the VIP project consisted of infants who had been admitted to the Neonatal Intensive Care Unit (NICU) of the University Medical Center Groningen between March 2003 and May 2005. At ten weeks corrected age (CA) a video recording was made of the infants' General Movements (GM). The assessment of the quality of GM can be considered as a technique to evaluate the quality of the brain function in young infants (Prechtl 1990, 2001, Hadders-Algra 2004).

General Movements

GM are spontaneously generated complex movements involving the head, trunk, arms and legs. They emerge at seven to eight weeks postmenstrual age and remain the most frequently observed movement pattern until the age of three to four months after term age when they gradually are replaced by goal-directed motor behaviour (Hadders-Algra 1996, 2001). Four classes can be distinguished in the assessment of GM quality: normal-optimal, normal-suboptimal, mildly abnormal and definitely abnormal GM. In typical development GM are characterized by a considerable variation and complexity and to a smaller extent by fluency. When GM are less optimal fluency is the first aspect that disappears, the movements become more jerky or stiff. Variation and complexity disappear when the movements become more abnormal. Various studies have shown that the presence of definitely abnormal GM at the age of two to four months corrected age (CA), the so-called fidgety GM phase, is associated with a high risk for developmental motor disorders such as CP (Prechtl et al 1997, Prechtl 2001, Hadders-Algra 2004, Groen et al 2005). Research showed that the GM method has a good predictive validity for both major and minor developmental disorders Heineman and Hadders-Algra, 2008). Hadders-Algra (2004) showed that the infants who show definitely abnormal GM, but do not develop a CP usually show other developmental problems, such as minor neurological dysfunction (MND) and behavioural problems. Also the construct and concurrent validity and reliability of the GM method are satisfactory (Heineman and Hadders-Algra 2008). The early detection of infants at high risk for developmental disorders offers the opportunity for intervention at young age, during a phase in which the central nervous system is characterized by high plasticity (Kolb et al 2001, De Graaf-Peters and Hadders-Algra 2006). Plasticity means that the brain can reorganize itself to some extent. Neurons neighbouring a lesioned part can change function and become incorporated into the affected repertoires, thereby increasing the variation of the movement repertoire of the child (Hadders-Algra 2000b).

During the period of March 2003 till May 2005 in total 257 video recordings of GM were made for the VIP project. Infants and their families were selected to participate in the project when the quality of the infants' GM was classified as definitely abnormal. Sixty-two infants showed definitely abnormal GM (Figure 2.1). Infants with severe congenital anomalies and infants whose caregivers had an inappropriate understanding of the Dutch language were excluded from the study. Forty-six families decided to participate in the VIP project. Through block randomization (full term infants blocks of n=2; preterm infants blocks of n=12) the infants were assigned to the experimental group who received the COPCA intervention (n=21) or to the control group who got the traditional form of paediatric physical therapy (TIP; n=25). The randomized intervention was provided between 3 to 6 months corrected age (CA). The COPCA intervention was carried out by 4 physiotherapists, who were intensively trained in the application of this method. COPCA was applied twice a week for one hour in a home based setting. The application of TIP varied. Infants received therapy with a median value of once a week, the mean duration of sessions was 30 minutes. TIP was also mainly applied in the home situation by the paediatric physiotherapist working in the area. After the intervention period physical therapy was only continued when the paediatrician in charge of the care of the infant considered it necessary. In the TIP group were three infants who did not receive physiotherapeutic guidance during the intervention period since their paediatrician did not refer them to a physiotherapist.

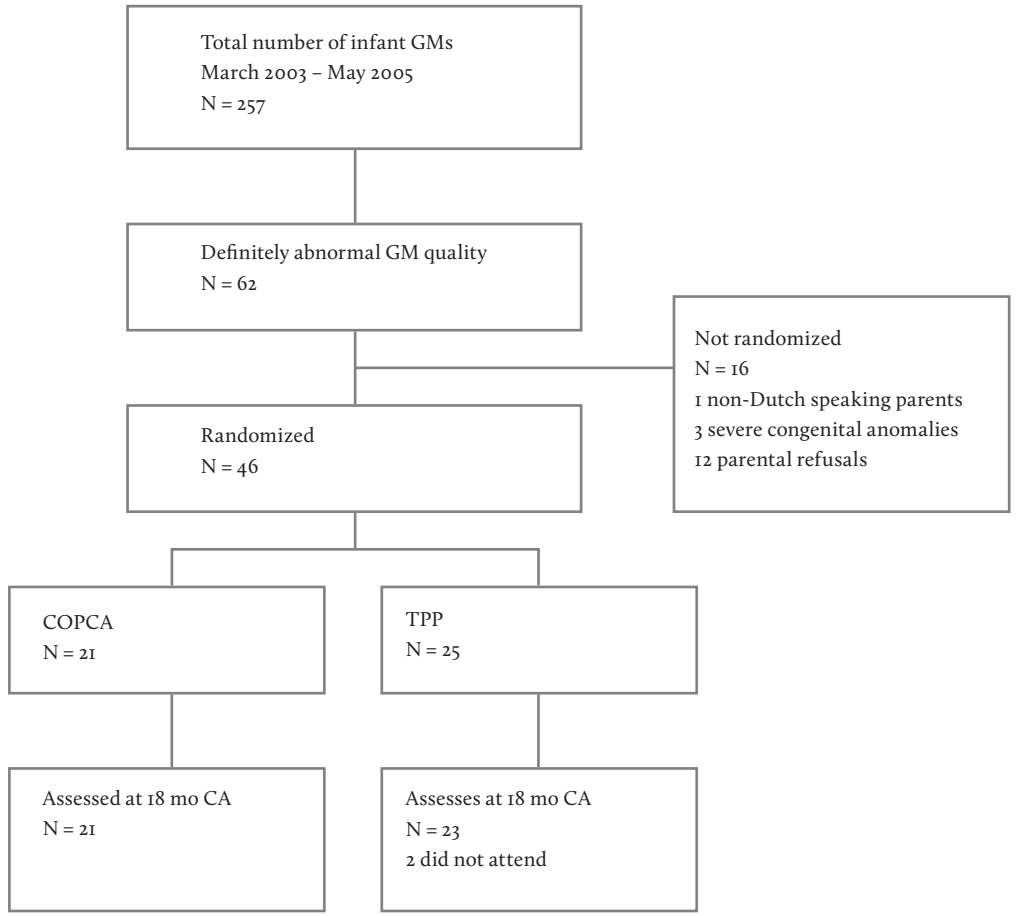

Figure 2.1. Infants included into the VIP project.

Evaluation and Outcome measures

The developmental status of the infants was assessed at 3, 4, 5, 6 and 18 months CA. Table 2.1 shows which outcome measures were assessed on the different evaluation moments. Neurological condition at 3 to 6 months was measured with the Touwen Infant Neurological Examination (TINE; Hadders-Algra et al 2009). At 18 months CA we used the neurological examination according to Hempel (1993). Additionally, at 18 months CA the concept of neurological optimality was applied to detect subtle differences in neurological condition (Huisman et al 1995). The Infant Motor Profile (IMP; Heineman et al 2008) was used to assess the neuromotor condition in a qualitative way. This instrument pays attention to the evaluation of motor abilities, movement variability, the ability to select motor strategies, movement symmetry, and fluency. Gross motor development was evaluated with the Alberta Infant Motor Scale (Piper and Darrah 1994). For the cognitive development of the infants the mental scale of the Bayley Scales of Infant Development was used (Bayley 1993, Van der Meulen et al 2002). During the assessments at 4, 6 and 18 months CA we also gathered information about the development of postural control of the infants.

Furthermore, we administered questionnaires to investigate the psychosocial development of the infants; the Kent Infant Development Scale (KIDS) (Schneider et al 1990, Reuter and Gruber 2000) and the Pediatric Evaluation of Disability Inventory (PEDI) (Haley et al 1992, Custers et al 2002a). Information about education of the infants was obtained with the Nijmegen Child-rearing Situation Questionnaire (NCSQ) (Wels and Robbroeckx 1996). The extent to which caregivers perceive the care they received as family centred and the contentment of the caregivers with the physiotherapeutic care was measured with the Measures of Processes of Care (MPOC; King et al 1995, van Schie et al 2004). The contents of the intervention and the application in daily life were evaluated by the use of video-analysis. Therefore we made videorecordings of intervention sessions at 4 and 6 months CA and recordings of activities of daily living (ADL) at 3, 6 and 18 months CA. A short description of the outcome measures is provided in this paragraph.

Table 2.1. Outcome measures.

3 months	4 months	5 months	6 months	18 months
TINE	TINE	TINE	TINE	Hempel + NOS
IMP	IMP	IMP	IMP	IMP
AIMS	AIMS	AIMS	AIMS	AIMS
			BSID –II	BSID- II
	Postural control		Postural control	Postural control
KIDS			KIDS	KIDS
PEDI			PEDI	PEDI
NCSQ			NCSQ	NCSQ
MPOC			MPOC	MPOC
	Video PT		Video PT	
Video ADL			Video ADL	Video ADL

AIMS = Alberta Infant Motor Scale, BSID-II = Bayley Scales of Infant Development, 2nd edition, IMP = Infant Motor Profile, KIDS = Kent Infant Development Scale, MPOC = Measures of Processes of Care, NOS = neurological optimality score, NCSQ = Nijmegen Questionnaire on Child-rearing Situations, PEDI = Pediatric Evaluation of Disability Inventory, TINE = Touwen Infant Neurological Examination, Video ADL = video of activities of daily living, Video PT = video of physical therapy session.

Neurological examination

To evaluate the neurological condition of the infants we used the Touwen Infant Neurological Examination (TINE; Hadders-Algra et al 2009). The neurological findings were summarized as normal, normal-suboptimal, minor neurological dysfunction (MND) or abnormal. During infancy the classification of abnormal implies the presence of a distinct neurological syndrome, such as a clear hypo- or hypertonia, a hemisyndrome, or a hyperexcitability syndrome. To distinguish between MND, normal-suboptimal and normal neurological condition, the findings of the TINE are classified according to age-specific norms into five clusters of dysfunction: dysfunctional reaching and grasping, dysfunctional gross motor function, brain stem dysfunction, visuomotor dysfunction and sensorimotor dysfunction. Two forms of normal neurological development can be distinguished: neurologically normal when none of the clusters meets the criteria for dysfunction and normal-suboptimal when one or two clusters fulfil the criteria for dysfunction. When more than two clusters fulfil the criteria for dysfunction infants are classified as MND. A recent study showed that MND can be assessed reliably with the TINE (Hadders-Algra et al 2009).

Neurological examination according to Hempel and Neurological Optimality Score (NOS)

The Hempel assessment is a standardized assessment technique designed for the detection of signs of minor neurological dysfunction during toddler age (1.5-4 years) (Hempel 1993). But of course, the assessment also allows for the evaluation of pathological conditions. Findings of the Hempel assessment were classified as neurologically normal, simple MND, complex MND or neurologically abnormal indicating conditions such as CP. The distinction between simple and complex MND is also based on the number of clusters of dysfunction. Simple MND at 18 months denotes the presence of one cluster of dysfunction and complex MND the presence of more than one cluster of dysfunction. The classification CP implies the presence of a 'classical' configuration of neurological signs (Hadders-Algra 2003).

Not only the traditional signs of neurological dysfunction, for example mild abnormalities in muscle tone or reflexes and motor milestones are assessed with the Hempel assessment. Through observation of spontaneous motor behaviour in a free field play situation the investigator can observe the qualitative aspects of spontaneous motor behaviour without compromising the cooperation of the child (Hempel 1993, Hadders-Algra 2005). The Hempel assessment consists of 94 items covering 7 sections including prehension function, sitting behaviour, crawling behaviour,

standing and walking behaviour, evaluation of cranial nerve functions, and sensorimotor function by means of manipulation including the assessment of muscle tone and the intensity and threshold of reflexes. Little information is available about the concurrent and predictive validity of the Hempel assessment. The interrater reliability is satisfactory (Hempel 1993).

Additionally at 18 months CA we used the Neurological Optimality Score (NOS) (Huisman et al 1995) to summarize neurological condition. Based on the neurological examination an optimality score can be calculated to detect small deviations in neurological condition. The NOS is the sum of 57 items representing the neurological examination that meet predefined criteria for optimality. Note that the definition of optimal is narrower than that of normal or typical, and that a reduced optimality not always means abnormal (Prechtl 1980). The NOS has been proven to be an excellent instrument to evaluate subtle differences in neurodevelopmental outcome (Bouwstra et al 2006).

Infant Motor Profile

The IMP is a video-based assessment of motor behaviour in infancy (Heineman et al 2008). The IMP consists of 80 items that evaluate motor abilities, movement variability, the ability to select motor strategies, movement symmetry, and fluency. It is applicable in children from 3 to 18 months. The advantage of the IMP is that it evaluates motor behaviour in a qualitative way. The IMP has sound psychometric properties. The intra- and interobserver agreement are satisfactory and the concurrent validity with the AIMS was good. Moreover the IMP was able to differentiate between infants with normal neurological condition, simple MND, complex MND, and abnormal neurological condition (Heineman et al 2008). No information is available on the predictive validity of the IMP.

Alberta Infant Motor Scale

The Alberta Infant Motor Scale (AIMS; Piper and Darrah 1994) is an observational instrument to measure gross motor development for infants from birth to independent walking. The AIMS consists of 58 items and is organized into four positions: prone, supine, sitting and standing. Through observation of spontaneous motor behaviour each item can be scored on three aspects of motor performance, namely weight bearing, posture and antigravity movements. The AIMS can be used for two purposes. First, to identify infants who are delayed or atypical in their motor performance and second to evaluate motor development over time (Piper and Darrah 1994). The AIMS has very good reliability coefficients (Piper and Darrah 1994, Heineman and Hadders-Algra 2008). Also the validity of the AIMS is well examined and proven to be satisfactory (Piper and Darrah 1994, Darrah et al 1998, Liao and Campbell 2004).

Bayley Scales of Infant Development

The Bayley Scales of Infant Development (BSID-II; Bayley 1993) is a psychometric sound instrument to assess mental and psychomotor development of children aged 1 to 42 months. In the early intervention project the Dutch version of the BSID-II was used to assess the cognitive development at the age of 6 and 18 months CA (Van der Meulen et al 2002). The BSID-II consists of a mental developmental index (MDI) and a psychomotor developmental index (PDI). A limitation of the BSID-II psychomotor scale is that the emphasis is placed on cognitive motor performances. Therefore we decided to administer only the MDI for the VIP project and to assess gross motor development with the AIMS and the qualitative aspects of motor performance with the IMP. The MDI was scored based on the items successfully completed. The MDI consists of items concerning problem solving, memory, discrimination, classification, language and social skills. Since the children were not exactly 6 or 18 months at time of the assessment, the scores were converted into age-equivalent scores, as derived from the Dutch norms (van der Meulen et al 2002). The interrater reliability of the MDI is sufficient, the construct and concurrent validity are moderate (Heineman and Hadders-Algra 2008).

Postural control

The development of postural control was assessed, because infants with high risk for developmental disorders often have problems with their postural control which interfere with the activities of daily life (Brogren et al 2001, Van der Heide et al 2004). Postural activity was assessed at 4 and 6 months CA during reaching while lying in supine and while sitting with support, and at 18 months CA while sitting with support and – when possible – without support. Reaching was elicited by presenting toys when the infant was in a calm and alert behavioural state. Elecromyografic (EMG) recordings were measured continuously during the testing session with bipolar surface electrodes placed on the following muscles: deltoid, pectoralis major, biceps brachii, triceps brachii, neck flexor (m. sternocleidomastoid), neck extensor, rectus abdominis, thoracal extensor, lumbar extensor, rectus femoris, and hamstrings on the right side of the body. Simultaneous with the EMG recordings, movements were recorded kinematically. For a detailed description of the procedure see de Graaf-Peters et al (2007).

Kent Inventory of Developmental Skills (KIDS)

The Kent Inventory of Developmental Skills is a caregiver-completed developmental assessment instrument for healthy infants, infants at risk, and for young children with developmental disabilities who are chronologically or developmentally under the age of 15 months (Schneider et al 1990, Reuter and Gruber 2000). The KIDS was used as comparison to the BSID-II. Both measures aim to evaluate the developmental level of the infant. At 3 months CA only the KIDS was administered to minimize the physical effort of the infant. At 6 and 18 months CA both instruments were assessed, being aware that the KIDS at 18 months might suffer from a ceiling effect. Parents were asked to fill out the questionnaire at home at 3,6, and 18 months CA and return it to the researchers.

The main idea of the KIDS is that all new behaviours are acquired in a more or less fixed order which is age dependent. The KIDS comprises of 252 behavioural items which are divided into five areas: motor behaviour (78 items), cognition (52 items), social behaviour (51 items), ability to

perform things independently (39 items) and language (38 items). Caregivers fill in whether or not their child shows the described behaviour. The total score provides an impression whether the developmental status of the child corresponds with the average chronological age of its peers with similar behavioural repertoires, and the status of the child as having no delay, at risk or developmentally delayed. Research showed that the interrater agreement, test-retest reliability and validity were high (Schneider et al 1990, Reuter and Gruber 2000). As the KIDS is a parent reported questionnaire it can be discussed whether caregivers are capable to objectively judge the development of their infant. Research has shown that most caregivers are very well capable of judging their infants' developmental level (Harris 1994, Johnson et al 2008). Therefore we assume that this is also the case in the VIP project.

Pediatric Evaluation of Disability Inventory

The Pediatric Evaluation of Disability Inventory (PEDI; Haley et al 1992) was used to measure the functional ability of the child. This assessment instrument is developed for young children from 6 months to 7.5 years of age with developmental disabilities and adapted to a Dutch version by Custers et al (2002a). The PEDI is a discriminative measure which aims to detect whether a child has a delay in his functional status and if so to determine the extent and content area of the delay. With the PEDI both the capability of the child (what a child can do) and the performance (what the child actually does do) of routine daily activities can be evaluated. Capability is measured by the functional skills scale (197 items), while the caregiver assistance scale and the modifications scale provide information on the performance (20 items). Each scale consists of three domains namely self-care, mobility and social function. The PEDI can be considered as a reliable and valid instrument (Custers et al 2002a, 2002b, Wassenberg-Severijnen et al 2003).

Measures of Processes of Care (MPOC)

The MPOC is designed to determine the extent to which caregivers perceive the health services they receive as family centred (King et al 1995, 1997). In the VIP project we used the Dutch translation of the MPOC (Van Schie et al 2004). Additionally, the MPOC was also used to evaluate whether the attitude of caregivers regarding health services changed over time. The MPOC is a 56-item self-administered questionnaire categorised in five domains: a) enabling and partnership, b) providing general information, c) providing specific information about the child, d) coordinated and comprehensive care, e) respectful and supportive care. The items ask to what extent a particular behaviour occurred during the past year. The responses are given on a 7-point scale ranging from 'not at all' to 'to a very great extent' with an additional 'not applicable' category. Domain scores are calculated as the mean of the ratings for the items in the scale and is only considered as valid when at least two third of the items in the scale received a score. The validity criterion for the questionnaire was completion of at least half of the items. The Dutch translation of the MPOC has sound psychometric properties; the construct validity and reliability measures are good (Van Schie et al 2004).

Nijmegen Child-rearing Situation Questionnaire (NCSQ)

The Nijmegen Child-rearing Situation Questionnaire (NCSQ, Wels & Robbroeckx 1996) was used to investigate the existing parenting situation. The questionnaire is based on the stress model of Lazarus and the attribution theory of Weiner (Wels and Robbroeckx 1996). The instrument consists of 4 scales: A) Subjective Family Stress. This scale consists of 46 items which form eight categories: a) acceptance of the child, b) coping, c) experience problems, d) need for change, e) child is a strain, f) managing on one's own, g) having pleasure, h) relation. B) Assessment of the childrearing situation. Parents are instructed to indicate which of eight descriptions fits best their own situation. In the first situation, no problems in child-rearing are present and support is absolutely unnecessary. In situation eight, child-rearing is highly problematic and parents are in urgent need of support. C) Attributions on the Child-rearing situation. This scale consists of 34 items which form five categories: a) ability, b) effort of oneself and partner, c) task difficulty, d) share of partner, e) luck/fate. D) Expectation for help. This last scale has 36 items divided over five categories: a) satisfaction, b) need for change, c) need for help, d) expectation for help (internal orientation), and e) expectation for help (external orientation). The answers of the y questions in scale A, C and D are rated on a 5-point Likert scale, ranging from 1 = "does not apply to me at all" to 5 = "applies to me". Sum scores were calculated for each category and converted into weighted scores. Higher scores indicate that the caregivers experience a relatively greater burden on the child's education. The reliability and the validity are sufficiently assessed (Wels and Robbroeckx 1996). Note however that later research showed that the norms are insufficiently assessed, because the reference groups were too small (Evers et al 2000). The caregivers are asked to fill in the NCSQ at 3, 6 and 18 months corrected age.

Video recordings of physical therapy sessions and ADL activities

During the VIP project video recordings were made of physiotherapeutic intervention sessions and of daily caregiving tasks. Albrecht et al (2005) showed that video recording hardly affects the behaviour of people that are being filmed, because they tend to forget the camera after a while. In order to ensure that the recording would resemble a natural situation and to minimize intrusion we took the following precautions: the camera was positioned as far away from the physiotherapist, caregiver and infant as possible and the physiotherapist and caregiver were asked to ignore the person making the video.

Video recordings of intervention sessions were made at 4 and 6 months CA. They were used to gain knowledge on the contents of paediatric physiotherapeutic intervention. On the one hand, this clearly showed whether the contents of COPCA were significantly different from the contents of TIP. On the other hand, the analysis of the video recordings enabled us to answer the question whether the COPCA programme could be well implemented in daily physiotherapeutic practice (see for more detail Chapter 5 and 6).

The video recordings of activities of daily living – i.e. bathing and playing – showed how well principles of the COPCA programme were implemented in the daily life of the families. These

activities were recorded at 3, 6 and 18 months CA. In this thesis the analysis and results of the ADL video recordings will not be further described.

The outcomes of the VIP project are presented in *Chapter 4, 5, 6, 7, and 8*. In *Chapter 4* some preliminary results of the first half of the study group on the neurological examination, NOS, AIMS and MDI of the BSID-II are described. *Chapter 5 and 6* report the results of the video analysis on the contents of paediatric physiotherapeutic intervention, i.e. COPCA and TIP. In *Chapter 7* the developmental outcome of the infants on the neurological examination, NOS, AIMS, MDI of the BSID-II and PEDI is described. Subsequently, to examine whether developmental outcome could be associated with treatment principles used by the PT or caregiver during intervention or with confounding factors we applied process evaluation to the data. Finally, in Chapter 8 an overview of the results on outcome measures which were also assessed during the VIP project, but were not the focus in the previous Chapters is provided. Attention is paid to the results of the following questionnaires: KIDS, MPOC, and NQCS. Also some preliminary results on the postural control of the infants are described. Data of the IMP, the ADL video recordings and postural control are currently being assessed.

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CHAPTER 3

A SYSTEMATIC REVIEW OF THE EFFECTS OF EARLY INTERVENTION ON MOTOR DEVELOPMENT

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Abstract

We present a systematic review on the effect of early intervention starting between birth and the corrected age of 18 months on motor development in infants at high risk for or with developmental motor disorders. Thirty-four studies fulfilled the selection-criteria.

Seventeen studies were applied within the NICU environment. Eight had a high methodological quality. They evaluated various forms of intervention. The results indicated that NIDCAP intervention might have a temporary positive effect on motor development. Twelve of the 17 post-NICU studies had a high methodological quality. They addressed the effect of NDT and specific or general developmental programmes. The results showed that intervention according to the principles of NDT does not have a beneficial effect on motor development. They also indicated that specific or general developmental programmes can have a positive effect on motor outcome.

We concluded that the type of intervention which might be beneficial for infants at preterm age differs from the type which is effective in infants who have reached at least term age. At preterm age infants seem to benefit most from intervention which aims at mimicking the intrauterine environment, such as NIDCAP intervention. After term age, intervention by means of specific or general developmental programmes has a positive effect on motor development.

Keywords

Early intervention, motor development, NDT, NIDCAP, infant stimulation

Introduction

In the last decades the importance of Early Intervention (EI) is generally recognized. But what exactly is early intervention? Typically a single definition is used, which is applied for EI to children at biological risk for developmental disorders and children with developmental disabilities. "Early Intervention consists of multidisciplinary services provided to children from birth to 5 years of age to promote child health and well-being, enhance emerging competencies, minimize developmental delays, remediate existing or emerging disabilities, prevent functional deterioration and promote adaptive parenting and overall family functioning. These goals are accomplished by individualized developmental, educational and therapeutic services for children in conjunction with mutually planned support for their families" (Shonkoff and Meisels 2000). In general, EI programmes use techniques derived from the domains of physiotherapy, occupational therapy, developmental psychology and education. Little attention is paid to the effect of nutrition, even though it is well known that cognitive outcome of breastfed children is significantly better than that of formula-fed children (Anderson et al 1999). The earliest studies on EI programmes primarily addressed the improvement in motor skills. Later, the focus shifted towards family-focused and other functional outcomes (Harris 1997). Thus, it appears that EI serves as an umbrella term covering the whole field of childhood intervention.

One of the problems associated with the use of the term EI is the interpretation of 'early'. 'Early' can be understood in two ways, namely as 'early in life' and as 'early in the expression of the condition'. Each of the two types of 'earliness' is associated with advantages and disadvantages for intervention. The major advantage of intervening early in life is that the brain is considered to be very plastic. The brain is especially plastic in the phase occurring after the completion of neuronal migration during which the processes of dendritic outgrowth and synapse formation are highly active (Kolb et al 2001). This means that high plasticity can be expected between 2 to 3 months before until about 6 to 8 months after term age (Hadders-Algra 2001). However, there are two potential disadvantages which might be associated with intervention early in life. First, the type of problems the infant will develop later in life will not yet be clear. This makes effective goal setting for EI difficult. Second, a part of the at-risk population will not develop a developmental disorder, thereby making intervention perhaps superfluous for these children.

Intervention in children with a developmental disorder in general starts later in life, i.e. at the time during infancy or preschool age when the condition is expressed in dysfunction. Two advantages of the latter situation are that intervention is applied to children who are really in need of EI and that the goals of the intervention can be formulated relatively easily. The most important disadvantage of intervention which first starts when the disorder has become undeniable is that it starts relatively late from a point of view of the plasticity of the brain (Hadders-Algra 2001). Indeed, previous studies indicated that programmes which started before the ninth month post term provided more improvement in both motor and personal skills than programmes starting later (Shonkoff and Hauser-Cram 1987, Sharkey et al 1990). Thus, it seems wise to start early in life.

Therefore, the aim of the present study is to systematically review studies on intervention starting early in life in children at high biological risk for developmental disorders. In particular we aim at unravelling the elements, which might contribute to a beneficial effect on motor development. Specific attention is paid to the timing of intervention in order to see whether we can find indications for the existence of a crucial age period in which EI results in the most beneficial outcome.

Method

Selection procedure

A literature search was performed using the following electronic databases: MEDLINE (1966 to July 2004), CINAHL (1982 to July 2004), AMED (1985 to July 2004), PsycINFO (1967 to July 2004) and PEDro. Reference lists in original studies and reviews were also examined for appropriate articles. The keyword 'early intervention' initially revealed 13,699 hits. In order to reduce the number of hits the following additional keywords were used: 'infant', 'motor development', 'low birth weight', 'preterm', 'high-risk' and 'cerebral palsy'. Thereafter the number of hits was limited to 485.

From these 485 papers only studies were included in the review when they fulfilled the following four criteria: a) subjects of study: infants with high biological risk for or with developmental disabilities, b) aim of the intervention (mostly amongst others): to improve motor development, c) onset of intervention in at least 50% of the participating children: between birth and the (corrected) age of 18 months, d) journal in which the study was published had an impact factor of > 0.3. Excluded from the review were studies restricted to medical and orthopaedic interventions and studies in populations of healthy low-risk preterm infants or in populations of socially disadvantaged children without specific biological risk for developmental disorders.

Based on the abstract, 60 papers were selected as potential candidates for the review. Reading the entire paper left 36 original studies which met all inclusion criteria. There were two studies within this selection of which the results were published in more than one paper (Resnick et al 1987, 1988, Palmer et al 1988, 1990). In the present review these studies were treated as single entities. The papers not included in the review mainly were review articles, studies focusing on improvement of cognitive function and studies in which the intervention was applied after the age of 18 months.

Evaluation procedure

The evaluation of the studies focused on the type and size of groups included in the study, the level of evidence (Table 3.1) and the internal and external validity of the study (criteria specified in Table 3.2). The level of evidence of a study and its internal and external validity were the determinants for the methodological quality of the study. All studies were rated independently by both reviewers. Interrater agreement was calculated for each of the determinants of methodological quality. Agreement was high: Cohen's Kappa varied from K = 0.86 (internal validity), K = 0.94 (level of evidence) to K = 0.95 (external validity). Disagreements were discussed till consensus was reached. In addition, attention was paid to specifics of the intervention programme, such as the type of intervention, its period of application and intensity, the location where the intervention had been carried out and parental involvement (Table 3.2). Finally, the age of the infants at evaluation, the outcome measures and the results were specified to further evaluate the effect of the intervention.

In order to assess the effect of the age period during which EI took place, we divided the studies into three age-groups (cf. Shonkoff & Hauser-Cram 1987, Sharkey et al 1990). The first group consisted of a) studies dealing with intervention programmes applied in the Neonatal Intensive Care Unit (NICU), b) the second group contained studies where the intervention programme started between discharge from NICU care and 9 months corrected age, c) the third group included studies where intervention started between 9 and 18 months corrected age.

The studies turned out to be so heterogeneous in type of intervention evaluated, outcome measures used to evaluate the effect of intervention and age at which outcome was studied, that no meta-analysis to examine effect sizes of interventions could be performed.

Table 3.1. Levels of evidence (Sackett 1989, Butler and Darrah 2001).

Level	*Group research*	*Single individual research*
I	Randomised controlled trials	N=1 randomised controlled trial
II	Non-randomised controlled trials	ABABA design
	Prospective cohort studies with concurrent control group	Alternating treatments (e.g. ABACA)
		Multiple baseline across participants
III	Case studies with control participants	ABA design
	Cohort studies with historical control group	
IV	Case series without control participants	AB design
V	Case reports	Case reports
	Non-empirical methods	Non-empirical methods

Results

Seventeen of the 34 studies dealt with interventions carried out within a NICU setting, eight evaluated intervention starting between discharge from the NICU and 9 months and six studies assessed intervention which started between 9 and 18 months corrected age. In three studies, the age at onset of intervention exceeded the pre-set period criteria. Therefore, a fourth category of age at onset of intervention was added, consisting of studies where intervention started at some age between discharge from the NICU and 18 months corrected age. Within the age periods, studies were ranked primarily according to level of evidence, next based on internal validity, external validity and effect (Tables 3.3-3.6).

Table 3.2. Evaluation criteria (modification of Siebes and Vermeer 2002).

Group & design		***Description***
Sample size		Total, Experimental group, control or contrast group
	?	Not present
Attrition		Number
	?	Unspecified
Study group		High-risk (i.e. preterm/LBW), CP, Down
	?	Unspecified
Levels of evidence		Levels according to Sackett's method for grading research (Sackett, 1989) modified by Butler and Darrah (2001); see Table 3.1.
Internal validity: can measured effects be attributed to intervention under study?	++	High internal validity
	+	Fair internal validity
	-	Low internal validity
		Internal validity can be reduced by various study variations such as subject assignment, contamination, co-intervention, and blind assessment
External validity: can results of research be generalized?	++	Generalization is plausible
	+	Some possibilities for generalization
	-	No information about generalization
Methodological quality		Based on level of evidence, internal and external validity
Intervention		
Method of treatment		
Contents description	++	Detailed description
	+	Summary only
	-	Very limited information
Period of application		Description
	?	Unspecified
Intensity		Description
	?	Unspecified
Location		Hospital, Home, Centre
		Combination
Parental involvement	P↑	Enhance parental skills
	T	Parent is therapist
	?	Unspecified
Evaluation of effect		
Age at evaluation		< term, around term, 1-9 mo post term, 9-18 mo, > 18 mo
Outcome measures		Neuromotor
		Developmental
Results		E=C, E>C, E<C

Populations and methods of intervention

The number of children included in the studies ranged from 10 to 746 individuals (median value: n= 44.5; Table 3.3). Information about the rate of attrition was provided in 33 studies. It varied from 0% to 64%, with a median value of 13%. In the majority of studies (24 out of 34) the study group consisted of so-called high-risk infants, i.e. infants born preterm or infants with a low birth weight. The remaining studies evaluated the effect of intervention in infants with cerebral palsy, delayed mental and motor development or Down syndrome.

The intervention programmes applied showed considerable diversity (Table 3.4). In 29 studies, at least brief information about the intervention method was given of which 10 studies gave a more detailed description. In order to get some insight into the intervention strategies and procedures applied, we assessed whether programmes contained the following elements: procedures to reduce stress, sensory stimulation (specific unimodal, specific multimodal, general multimodal), motor intervention strategies (passive handling techniques, active training of specific motor abilities, general motor training) and parent infant interaction strategies (Table 3.6). Both authors assessed the composition of the intervention programmes independently. Interrater agreement was high: Cohen's Kappa for the various components varied from 0.79 to 1.00. NICU intervention programmes mainly consisted of combinations of procedures aiming at the reduction of stress, the provision of auditory, tactile, visual or vestibular stimuli and passive motor handling procedures. Amongst the programmes which started after discharge from the NICU, Neurodevelopmental Treatment (NDT), which consists of a mix of general sensory stimulation and passive and active motor intervention strategies, was the intervention most frequently used. Other frequently applied forms of intervention were various forms of developmental programmes, which always included general sensory stimulation and general stimulation of motor development, but also could imply passive handling techniques and the enhancement of parent infant interaction.

Information about the period of application of the intervention was supplied in 30 studies. Most NICU interventions were applied during variable periods, as most interventions took place between the age of some postnatal days till discharge. The application period in the post-NICU studies varied between 2 months and more than 4 years. Also the intensity of intervention showed considerable heterogeneity. It ranged from once a month to continuous intervention. Programmes starting after discharge from the NICU were mostly applied within a home based setting. Another frequently used way to supply intervention was the use of hospital or centre based intervention combined with home care. In 24 out of the 34 studies parents were incorporated into the intervention. In 19 studies the focus was on enhancing parental skills, in the other five studies the parents carried out (a part of) the intervention, i.e. the parents functioned as a therapist.

General evaluation of effects (Tables 3.3-3.7)

Most studies evaluated the effect of intervention on motor performance during the intervention, immediately after the end of intervention and/or some months or 1-2 years after intervention. Various sorts of outcome measures were used for effect evaluation. We categorized the measures into

Table 3.3. Groups and design.

Study	*N*	*E*	*C*	*Attrition (%)*	*Study group*	*Evidence level*	*Internal validity*	*External validity*
NICU								
Als et al. 1994	38	20	18	0	HR	I	++	++
Resnick et al. 1987/1988	255	107	114	13	HR	I	+	+
Darrah et al. 1994	107	53	54	51	HR	I	+	+
Ariagno et al. 1997	35	14	14	20	HR	I	+	+
Charpak et al. 2001	746	382	364	16	HR	I	+	+
Nelson et al. 2001	37	21	16	30	HR or CNS injury	I	+	+
Kleberg et al. 2002	25	11	9	20	HR	I	+	+
Tessier et al. 2003	431	183	153	22	HR	I	+	+
Westrup et al. 2004	41	21	20	37	HR	I	+	-
Korner et al. 1983	56	12	8	64	HR	I	-	-
Feldman et al. 2002	146	73	73	9	HR	II	-	-
Helders et al. 1989	149	67	82	34	HR	II	-	-
Als et al. 1986	16	8	8	0	HR	III	+	-
Leib et al. 1980	28	14	14	0	HR	III	-	-
Mouradian and Als 1994	40	20	20	0	HR	III	-	-
Becker et al. 1999	38	?	?	61	HR	III	-	-
Kleberg et al. 2000	33	15	18	?	HR	III	-	-
After NICU – 9 mo								
Goodman et al. 1985	80	40	40	0[1]	HR	I	++	++
Piper et al. 1986	134	66	68	14	HR	I	++	++
Leksculchai and Cole 2001	84	43	41	14	HR	I	++	+
Barrera et al. 1986	80	32	48	26	HR	I	++	+
Weindling et al. 1996	105	51	54	21	HR of CP	I	++	+
Rothberg et al. 1991	49	28	21	39[1]	HR	I	+	+
D' Avignon et al. 1981	32	12/10[2]	8	6	HR	I	+	-
Kanda et al. 2004	10	5	5	0	CP	III	-	-
9 mo – 18 mo								
Ulrich et al. 2001	30	15	15	0	DS	I	++	++
Palmer et al. 1988/1990	48	25	23	2	CP	I	++	++
Reddihough et al. 1998	66	32	34	9	CP	I	+	+
Mahoney 2001	50	28	22	0	CP/ DS	II	++	+
Eickmann et al. 2003	156	78	78	13	Delayed cogn/ mot dev	II	+	+
Piper and Pless 1980	37	21	16	0	DS	III	+	-
After NICU – 18 mo								
Mayo 1991	29	17	12	0	Suspected CP	I	++	+
Scherzer et al. 1976	24	14	8	8	CP	I	++	+
Harris 1981	20	10	10	0	DS	I	+	+

C = Control/contrast group, CNS = Central Nervous System, CP = cerebral palsy, Delayed cogn/mot dev = Delayed cognitive/motor development, DS = Down syndrome, E = Experimental group, HR = High risk

[1] In the time between the two studies 31 children were lost to follow-up

[2] Ten infants received a different kind of intervention

neuromotor ones or as tests which provide a more general description of the child's developmental level (Table 3.7). The Bayley Scales of Infant Development were most frequently used as outcome measure for both neuromotor (PDI) and developmental (MDI) outcome. In 26 studies developmental tests were used to evaluate the effect of intervention. In nine of these a beneficial effect of intervention on the developmental parameters was reported. In 26 studies neuromotor tests were used. In 13 of these, experimental infants had a better neuromotor outcome than control infants.

Most studies were designed as randomised controlled trials. Twenty-three of the 34 studies had the highest level of evidence, i.e. level I according to Sackett (see Table 3.1), four studies had a grade II level of evidence and seven studies were classified as level III. Internal validity was high in 11 studies, fair in 15 and low in eight studies. External validity was in general moderate only: in five studies generalization was plausible, 17 studies offered some possibilities for generalization and 12 studies had low external validity. Validity of post-NICU studies was usually better than that of NICU studies.

In the next paragraphs we report the effects of intervention on motor development for the different age periods during which intervention had started, while taking into account the methodological strength of the studies.

Effect of intervention starting in the NICU

From the 17 NICU studies, eight had a high methodological quality. These studies had an evidence level I, a fair to high internal validity and they provided at least some possibilities for generalization. In two of these eight studies a significantly positive effect of intervention on motor outcome could be demonstrated. One of the positive studies dealt with the effect of NIDCAP (Als et al 1994). The focus of intervention in this study was stress reduction in combination with general sensory stimulation. The other intervention aimed at improving the infant's general developmental level by means of a developmental programme, including general sensory stimulation, general stimulation of motor development, passive handling techniques and the enhancement of parent infant interaction (Resnick et al 1987, 1988). In the other six NICU studies of high methodological quality, intervention had no statistically significant effect on motor development (Darrah et al 1994, Nelson et al 2001, Ariagno et al 1997, Charpak et al 2001, Kleberg et al 2002, Tessier et al 2003). The interventions used in these studies all included procedures to reduce the infant's level of stress and multimodal sensory stimulation (either specific or general), which was or was not combined with passive motor intervention techniques or the facilitation of parent infant interaction.

Five of the nine NICU studies with a lower methodological quality pointed to a positive effect of intervention (Als et al 1986, Mouradian and Als 1994, Feldman et al 2002, Korner et al 1983, Leib et al 1980). Intervention used in these studies consisted of various combinations of procedures to reduce stress, multimodal sensory stimulation, passive motor intervention strategies or the facilitation of parent infant interaction. The remaining four studies were unable to demonstrate a beneficial effect of intervention (Kleberg et al 2000, Westrup et al 2004, Helders et al 1989, Becker et al 1999).

Effect of intervention starting between NICU-discharge and 9 months

Six of the eight studies in which intervention started in the period between discharge from the NICU and the corrected age of 9 months had a high methodological quality. In four studies, amongst them the two methodologically strongest studies, the effects of NDT were evaluated (Goodman et al 1985, Piper et al 1986, Rothberg et al 1991, Weindling et al 1996). None of the NDT-studies demonstrated a statistically significant effect of intervention on motor development. It should be noted however, that in two studies (Goodman et al 1985, Rothberg et al 1991) outcome was evaluated only by means of global developmental tests. Two other studies (Barrera et al 1986, Leksculchai and Cole 2001) applied a programme to stimulate infant motor development. Both programmes consisted of general sensory stimulation and general stimulation of motor development. In addition, the intervention in the Barrera et al. study included enhancement of parent infant interaction, that of Leksculchai and Cole passive handling techniques. Both studies reported a statistically significant positive effect of intervention on motor development. Two studies addressing the effect of intervention starting in this age period were of limited methodological quality. One study evaluated in relatively small groups the effect of NDT and that of treatment according to Vojta (d'Avignon et al 1981). It concluded that outcome in the two treatment groups did not show a statistically significant difference. The other study reported a dose-response effect of treatment according to Vojta on motor outcome (Kanda et al 2004).

Effect of intervention starting between 9 and 18 months

Three of the six post-NICU studies, in which intervention started between 9 and 18 months of age, had a high methodological quality. One study demonstrated a positive effect of specific motor training on motor development of infants with Down syndrome (Ulrich et al 2001), another revealed that a general infant stimulation programme facilitated motor development more than NDT did (Palmer et al 1988, 1990). The third did not find a significant effect of conductive education on motor development (Reddihough et al 1998). The other three late post-NICU studies had a more limited methodological quality. One evaluated the effect of NDT and did not demonstrate a significant positive effect of intervention (Mahoney 2001). The other two used general programmes to stimulate motor development. One of them found a beneficial effect of intervention (Eickmann et al 2003).

Effect of intervention starting between NICU-discharge and 18 months

The three studies in which intervention started between discharge from the NICU and the age of 18 months were all level I studies with a moderate to high internal validity and a moderate external validity. Two studies evaluated the effect of NDT. The study which assessed the effect of a short period of intensive NDT was unable to demonstrate a significant effect of intervention on motor development (Harris 1981), the other which applied less intensive NDT for half a year did report a positive effect of intervention on motor development (Mayo 1991). The third study evaluated the effect of a general physical therapy programme and did not find a significant beneficial effect of the intervention on motor development (Scherzer et al 1976).

Table 3.4. Type of intervention.

Study	Method	Descr.	Period of application		Intensity	Location	Parents
			Onset	End			
NICU							
Als et al. 1994	NIDCAP	++	3 d after birth	?	Continuously	Hp	P↑
Resnick et al. 1987/1988	Dev Int	++	Birth	24 mo	Hp: daily Hm: 2x/mo	Hp/Hm	P↑
Darrah et al. 1994	Wb	+	2-7 d after birth	Placement in open cots	Continuously	Hp	?
Ariagno et al. 1997	NIDCAP	+	?	?	Continuously	Hp	P↑
Charpak et al. 2001	KC	+	3-5 d after birth	Position no longer accepted	Continuously	Hp	P↑
Nelson et al. 2001	ATVV	++	33 wk PMA	2 mo	Hp: 15 min, 2x/d 5x/wk , Hm: 2x/d	Hp/Hm	T
Kleberg et al. 2002	NIDCAP	+	1 d after birth	?	Continuously	Hp	P↑
Tessier et al. 2003	KC	++	condition stable	Discharge	Continuously	Hp	P↑
Westrup et al. 2004	NIDCAP	+	1 d after birth	36 wk PMA	Continuously	Hp	P↑
Korner et al. 1983	Wb	+	< 4 d after birth	Till evaluation	Continuously	Hp	?
Feldman et al. 2002	KC	++	31-34 wk PMA	Discharge, duration ≥ 14d	> 1 h/d	Hp	P↑
Helders et al. 1989	T stim/ RF	-	Birth	Discharge	?	Hp	?
Als et al. 1986	NIDCAP	++	9 d after birth	Discharge	Continuously	Hp	P↑
Leib et al. 1980	Sens Enr	+	Birth	Discharge	?	Hp	?
Mouradian and Als 1994	NIDCAP	+	?	< 40 w PMA	Continuously	Hp	P↑
Becker et al. 1999	Dev hand	+	Birth	36 wk PMA	?	Hp	?
Kleberg et al. 2000	NIDCAP	+	3 d after birth	36 wk PMA	?	Hp	P↑
After NICU – 9 mo							
Goodman et al. 1985	NDT	-	3 mo	12 mo	I: 1x/mo > 45 min P: daily (home programme)	Hp/Hm	P↑
Piper et al. 1986	NDT	+	Term age	12 mo	I: 0-3 mo: 1x/wk 3-12 mo: 1x/2wk P: daily	Hm	P↑
Leksculchai and Cole 2001	Dev pgm	++	Term age	4 mo	I : 1x/mo P: daily	Hm	T
Barrera et al. 1986	DPI /PIT	+	4 mo	16 mo	I: 4-7 mo: 1-2 h/wk 7-13 mo: 1x/2wk 13-16 mo: 1x/mo	Hm	P↑
Weindling et al. 1996	NDT	-	Term age	12 mo (CP → continue)	I: 0-6 mo: 1x/wk 6-9 mo: 1x/2wk 9-12 mo: 1x/mo	Hm	?
Rothberg et al. 1991	NDT	+	3 mo	12 mo	I: 1x/mo > 45 min P: daily (home programme)	Hp/Hm	P↑
D' Avignon et al. 1981	Vojta/NDT	-	4-7 mo	B: > 3 mo V: > 6 mo	?	?	?
Kanda et al. 2004	Vojta	-	1 mo	E: Mean 52 mo C: Mean 25 mo	E: ≥ 30 min, 3-4x/day	Hm	T
9 mo – 18 mo							
Ulrich et al. 2001	TT	+	9-12 mo	Independent walking	8 min/d, 5 d/wk	Hm	T
Palmer et al. 1988/1990	NDT/ IS	++	12-19 mo	Duration 12 mo	I: 1 h 1x/2wk P: daily (home programme)	Ce/Hm	P↑
Reddihough et al. 1998	CE	++	12-36 mo	Duration 6 mo	E: 2.8 h/wk C: 2.9 h/wk	Ce	?
Mahoney 2001	NDT/ DevS	+	Mean age 14 mo	Duration 12 mo	3x/mo, 45 min	Ce/Hm	?
Eickmann et al. 2003	Dev stim	+	13 mo	18 mo	I: 11 hm visits 30-45 min, 3 workshops	Hm	P↑
Piper and Pless 1980	Dev mile	+	Mean age 9 mo	Duration 6 mo	I: 1 h 1x/2wk P: home programme	Ce/Hm	P↑
After NICU – 18 mo							
Mayo 1991	NDT	+	4-18 mo	Duration 6 mo	E: 1 h/wk C: 1 h/mo	Hp/Hm	P↑
Scherzer et al 1976	Phys T	+	5-17 mo	24 mo	1h 1x/2wk	Hm	T
Harris 1981	NDT	++	2-21 mo	Duration 9 wk	E: 3x/wk >40 min	Hm	?

ATVV = Auditory-Tactile-Visual-Vestibular Stimulation, C = Control group, Ce = Centre, CE = Conductive Education, d = day, Descr. = description, Dev hand = Developmental Handling, Dev Int = Developmental Intervention, Dev mile = Developmental milestones, Dev pgm = Developmental programme, Dev S = Developmental Skills, Dev stim = Developmental stimulation, DPI = Developmental Program Intervention, E = Experimental group, h = hour, Hm = home, Hp = Hospital, I = Instruction, IS = Infant Stimulation, KC = Kangaroo Care, min = minutes, mo = months (corrected age), NDT = Neurodevelopmental Treatment, NIDCAP = Newborn Individualized Developmental Care and Assessment Program, P = parents, P↑= enhance parental skills, PIT = Parent-Infant treatment, PMA = post menstrual age, Phys T = Physical Therapy, Sens. Enr. = Sensory Enrichment, T = parents are 'therapist,' T stim/RF = Tactile Stimulation/ Range Finding, TT = Treadmill Training, Wb = Waterbed, wk = weeks, ? = no information available

Table 3.5. Evaluation of effect of intervention.

	Ages of evaluation					Outcome measures[1]		Results[2]
	<Term	Term	1-9 mo	9-18 mo	>18 mo	Neuromotor	Developmental	
NICU								
Als et al. 1994	-	-	1	9	-	APIB, Bayley PDI	Bayley MDI	E > C
Resnick et al. 1987/1988	-	-	-	12	24	Bayley PDI	Bayley MDI	E > C
Darrah et al. 1994	1x	+	4,8	12	18	MAI	Peabody	E = C
Ariagno et al. 1997	1x	-	1,4	12	24	Bayley PDI, NAPI, APIB	Bayley MDI	E = C
Charpak et al. 2001	-	+	3,6	9,12	-	-	Griffith	E = C
Nelson et al. 2001	-	-	2,4	12	-	Bayley PDI	Bayley MDI	E = C
Kleberg et al. 2002	-	-	-	12	-	Bayley PDI	Bayley MDI	E = C
Tessier et al. 2003	-	-	-	12	-	-	Griffith	E = C
Westrup et al. 2004	-	-	-	-	66	Movement ABC	-	E = C
Korner et al. 1983	1x	-	-	-	-	LAPPI	-	E > C
Feldman et al. 2002	1x	-	3,6	-	-	Bayley PDI	Bayley MDI	E > C
Helders et al. 1989	pre, after 2wk, weekly till discharge				-	-	PM Dev	E = C
Als et al. 1986	4x	+	1,3,6	9	-	APIB, Bayley PDI	Bayley MDI	E > C
Leib et al. 1980	2x	-	6	-	-	Bayley PDI, NBAS	Bayley MDI	E > C
Mouradian and Als 1994	-	-	1	-	-	APIB	-	E > C
Becker et al. 1999	3x	-	-	-	-	VA	-	E = C
Kleberg et al. 2000	-	-	-	-	36	-	Griffith	E = C
After NICU – 9 mo								
Goodman et al. 1985	-	-	6	9,12	-	-	Griffith	E = C
Piper et al. 1986	-	-	6	12	-	Wolanski	Milani, Griffith	E = C
Leksculchai and Cole 2001	-	+	1,2,3,4	-	-	TIMP	-	E > C
Barrera et al. 1986	-	-	4	16	-	Bayley PDI	Bayley MDI	E > C
Weindling et al. 1996	-	-	-	12	30	MAI	Griffith	E = C
Rothberg et al. 1991	-	-	-	-	72	-	Griffith-II	E = C
D'Avignon et al. 1981	-	-	-	-	36-72	CP class	-	E = C
Kanda et al. 2004	-	-	1,2,3,5	every 3 mo	59	NeurEx	-	E > C
9 mo – 18 mo								
Ulrich et al. 2001	-	-		every 2 wk		Bayley PDI	Bayley MDI	E > C
Palmer et al. 1988/1990	-	-	-	-	18,24	Bayley PDI	Bayley MDI	E < C
Reddihough et al. 1998	-	-		pre-post		GMFM	-	E = C
Mahoney 2001	-	-	-	-	14,26	-	Bayley MDI, Peabody	E = C
Eickmann et al. 2003	-	-	-	12	18	Bayley PDI	Bayley MDI	E > C
Piper and Pless 1980	-	-	-	pre-post		-	Griffith	E = C
After NICU – 18 mo								
Mayo 1991	-	-	pre		post	Wolanski	Bayley	E > C
Scherzer et al 1976	-	-	pre		24	MDC	MDI,Gesell Gesell	E = C
Harris 1981	-	-	pre		post	Bayley PDI	Bayley MDI, Peabody	E = C

C = Control group, E = Experimental group, mo = months, MP = motor performance, pre = before intervention, post = after intervention, wk = weeks

[1] For explanation of abbreviations see Table 3.7

[2] Statistically significant differences found in motor outcome at oldest age of evaluation: E > C: Experimental group significantly better outcome than Control Group, E = C: no difference between groups, E < C: Control Group better outcome than Experimental Group.

Table 3.6. Composition of programmes used in EI.

Intervention		Sensory Stimulation			Motor Intervention			Enhancing PII
	S. R.	S. u.	S. m.	G. m.	P.	A. s.	A. g.	
Waterbed	+	-	+	-	+	-	-	-
ATVV intervention	+	-	+	-	-	-	-	-
NIDCAP	+	-	-	+	+	-	-	+
KC	+	-	-	+	+	-	-	+
Developmental handling	+	-	-	+	+	-	-	?
Treadmill training	-	+	-	-	-	+	-	-
Tactile stimulation/Range finding	-	+	-	-	+	-	-	-
Vojta	-	+	-	-	+	-	-	-
Sensory enrichment	-	-	+	-	-	-	-	-
Developmental intervention	-	-	+	-	+	-	+	+
NDT	-	-	-	+	+	-	+	-
Developmental program	-	-	-	+	+	-	+	-
Developmental milestones	-	-	-	+	-	-	+	+
Developmental Stimulation	-	-	-	+	-	-	+	+
Developmental Parent intervention	-	-	-	+	-	-	+	+
Infant Stimulation	-	-	-	+	-	-	+	+
Conductive Education	-	-	-	+	-	-	+	-
Developmental skills	-	-	-	+	-	-	+	?
Parent Infant treatment	-	-	-	-	-	-	-	+

+ = procedure used, - = procedure not used,? = insufficient information available,
PII = Parent Infant Interaction; for other abbreviations see legends Table 3.4.

S. R. = Stress Reduction, S. u. = Specific –unimodal, S. m. = Specific – multimodal, G. m. = General – multimodal, P. = Passive, A. s. = Active – specific, A. g. = Active – general

Stress reduction: decreasing stressful events to the body by restricting input from the environment until the infant is capable of maintaining an adequate organization of its behavioural state; placing the infant in a way as to provide a sense of containment similar to the intrauterine environment.

Sensory stimulation:

a) specific-unimodal: procedures during which a single sensory modality is stimulated (e.g., specific tactile stimulation)

b) specific-multimodal: procedures during which multiple specific sensory modalities are stimulated (e.g., ATVV which consists of the application of auditory, tactile, visual and vestibular stimuli)

c) general-multimodal: procedures during which multiple forms of not explicitly described sensory stimuli are applied (e.g., verbal and tactile encouragement as part of general developmental programmes)

Motor intervention strategies:

a) passive procedures: the therapist or the parent carries out specific techniques, which do not require active motor behaviour of the child, i.e., the child has a passive role (e.g. handling, positioning, and facilitation procedures)

b) active-specific: the child is encouraged to actively train a specific motor ability (e.g., walking by means of treadmill training)

c) active-general: the child is encouraged to train a variety of motor abilities; stimulation of activities occurs in general by means of structured activities which are designed to meet the child's developmental level. Practice and play are important elements in the latter type of intervention.

Enhancing parent-infant interaction: parents receive information on infant behaviour. The increased knowledge on infantile behaviour facilitates parent's sensitivity to the child's needs and promotes developmentally supportive behaviour.

Discussion

We are not the first to write a review on the effects of early intervention for children at high risk for developmental disabilities, others preceded us: Simeonsson et al 1982, Parette and Hourcade 1984, Casto and Mastropieri 1986, Shonkoff and Hauser-Cram 1987, Parry 1992, Turnbull 1993, Guralnick 1997, Majnemer 1998. The major conclusion from these reviews is that the evidence favouring EI is inconclusive. The results from this present review indicate that we have moved a bit forward. In the following paragraphs we will point out the direction of progress. But before we address the issues of 'which programme at what age', we first discuss some methodological issues.

Methodological considerations

The studies included in this review were very heterogenic in nature. Not only a large variation existed in the number of subjects included in the studies, but also in the intervention methods which started prior to 18 months. The interventions themselves, the outcome measures used to evaluate the intervention methods and the ages at which outcome was determined were too heterogeneous to allow for a formal meta-analysis. The methods of intervention in the NICU period varied from NIDCAP and Kangaroo Care to different kinds of stimulation programmes. In post NICU studies NDT was the leading method of intervention, but there were also studies which used interventions like infant stimulation, conductive education and other developmental programmes.

It was encouraging to notice that 20 out of 34 studies had a high methodological quality, i.e., they had an evidence level I and a fair to high internal and external validity. This reflects that during recent years the requirement of good quality studies on the effect of early intervention has been increasingly materialized (Siebes et al 2002). Previous studies indicated that with an increase in the rigorousness of the studies the support for effectiveness of EI decreases (Parette and Hourcade 1984, Siebes et al 2002). For the studies included in the present review this also holds true. Of the 20 studies with a high methodological quality only six (30%) were able to demonstrate a sig-

Table 3.7. Classification of outcome measures into neuromotor tests and developmental tests.

Neuromotor test			**Developmental test**		
Bayley Scales of Infant Development - Psychomotor Development Index	Bayley-PDI	1969 1993	Griffiths Developmental Scales	Griffith	1954 1970
Wolanski Gross Motor Evaluation	Wolanski	1973	Milani-Comparetti Motor Development Screening test	Milani	1967
Neonatal Behavioral Assessment Score	NBAS	1973 1984	Bayley Scales of Infant Development - Mental Development Index	Bayley-MDI	1969 1993
Motor Development Checklist	MDC	1976	Gesell Developmental Schedules	Gesell	1974
Movement Assessment of Infants	MAI	1980	Peabody Developmental Motor Scales	Peabody	1974
Assessment of Preterm Infants' Behavior	APIB	1982	Muenchener Funktionelle Entwicklungs Diagnostik	MFED	1978
Longitudinal neurobehavioral assessment procedure for preterm infants	LAPPI	1983	Psychomotor Development profile (based on Gesell)	PM Dev	1981
Supplemental Motor Test for Postural Control	PoCo	1987			
Neurobehavioral Assessment of the PretermInfant	NAPI	1990			
Movement Assessment Battery for Children	Movement ABC	1992			
Gross Motor Function Measure	GMFM	1993			
Test of Infant Motor Performance	TIMP	1995			
Video-analysis (amount and variation in movements)	VA	1999			
Clinical Neurological examination	Clin Neur Ex	n.f.s.*			
CP classification	CP class	1981			

*not further specified

nificant beneficial effect of intervention on motor development. From the 14 studies with a limited methodological quality, seven (50%) reported a positive effect of intervention.

Studies on the effect of intervention in children with or at risk for developmental disorders - like the ones included in the present review - are often hampered by specific problems (Majnemer 1998, Siebes et al 2002). First, many studies include small study groups with a large heterogeneity of degree and type of problems thereby diminishing generalizability and statistical power. Second, assigning participants to a control group which does not receive treatment usually is considered unethical. Therefore, the results of most intervention studies represent only the additional value of the intervention under study. Another significant problem concerns the existing standardized outcome measures. In general they are characterized by a lack of sensitivity to detect small changes in motor development although these small changes may have an important influence on the functional abilities of the child. The studies included in this review not only used a large variation in outcome variables (see Table 3.7), but the measures were also mainly discriminative. The use of discriminative measures, which focus on the comparison of a child's score with an age-equivalent score, may be one of the reasons that so little effect of EI is found. Another problem associated with the used outcome measures is that they mainly measure quantitative changes in motor development instead of qualitative ones and measures which focus on changes in functional abilities (Ketelaar et al 1998).

Few studies addressed the effect of intervention on outcome beyond preschool age. This means that we lack information on the effect of EI on the child's activities of daily life and the child's socialization brought about by the potentially beneficial effect of EI on motor development. Future studies should address the effect of EI on these outcome parameters, as they have a major impact on the child's participation in society.

The results of our review are discussed while taking age at the onset of intervention as a primary focus. The number of studies with a high methodological quality in the various age periods after NICU discharge was so low that it precluded conclusions on the effect of age at onset of intervention after term age. We therefore decided to discuss the results of our review in two sections, one for NICU studies and one for post-NICU studies.

NICU studies

Eight of the 17 NICU studies had a high methodological quality. Three of those evaluated the effects of NIDCAP intervention. One demonstrated a significant positive effect on motor development as measured by the psychomotor index of the Bayley Scales of Infant Development (Bayley - PDI; Als et al 1994), but the other two were unable to reveal such an effect on Bayley – PDI (Kleberg et al 2002, Ariagno et al 1997). The difference in outcome between the three studies might be attributed to the age at which outcome was assessed.

Outcome in the positive effect study of Als et al (1994) was evaluated at 9 months corrected age, in the two 'no effect' studies at 12 and 24 months. Thus, it could be that NIDCAP has a temporary beneficial effect on motor development of infants at high risk for developmental disorder.

This notion is in line with results of two recent meta-analyses which concluded that NIDCAP has a temporary beneficial effect on cognitive and motor development (Jacobs et al 2002, Symington and Pinelli 2003). But it should be kept in mind that only the study of Westrup et al (2004) assessed the effect of NIDCAP beyond the age of two years. The latter small study was unable to demonstrate a significant positive effect of NIDCAP on developmental outcome at the age of 5½ years. Nevertheless, considering reports that NIDCAP intervention in low risk preterm infants has a significant positive effect on electrophysiological and MRI correlates of brain development at 42 weeks postmenstrual age (Buehler et al 1995, Als et al 2004), it is conceivable that NIDCAP might affect complex motor behaviour and cognitive abilities at school age. It might be that this putatively positive effect will be found in particular in low risk preterm infants and not in high risk preterms.

Two other high quality studies used Kangaroo Care to improve motor outcome (Charpak et al 2001, Tessier et al 2003). The application of Kangaroo Care had no effect on developmental outcome as measured by the Griffiths Developmental Scales at 6 and 12 months corrected age. Two explanations for this result can be offered. First, it could be that Kangaroo Care does not affect motor development. It is likely that the effect of the relatively simple Kangaroo Care is weaker than that of the rather complex NIDCAP programme. Second, it is possible that the effects are too subtle to be caught by the Griffiths scales.

Two high quality studies applied intervention strategies consisting of procedures to reduce stress in combination with specific multimodal sensory stimulation with or without passive motor intervention procedures (Darrah et al 1994, Nelson et al 2001). Both studies were unable to find a positive effect of intervention on motor development at 12 and 18 months corrected age.

The last high quality NICU study showed that developmental intervention had a significant positive effect on motor development (Resnick et al 1987, 1988). In this programme intervention started in the hospital with vestibular and visual stimulation to promote development. After discharge, intervention was continued for two years by means of a developmental programme in the home situation. The parents played an important role in carrying out the intervention programme which consisted of 400 different motor, social and cognitive activities complemented by several parenting activities. The positive outcome of the intervention probably can be attributed more to the duration of the programme and the continuous involvement of the parents in the development of their children than by the NICU part of the intervention.

In conclusion, the current review provides little evidence that intervention during the NICU period in infants at high risk for developmental disorders has a beneficial effect on motor development. Yet, a potential advantageous effect of NIDCAP on motor development cannot be excluded. We recommend that further studies address the effect of NIDCAP on developmental outcome at school age in low risk and high risk preterm infants.

Post NICU studies

From the 17 studies which started after the NICU period, 12 had a high methodological quality. Only four of those were able to show a beneficial effect of intervention on motor development. Eight of the 12 studies evaluated the effects of NDT or physiotherapy mainly based on the principles of NDT. It is striking that only one of these studies reported a better motor outcome in the experimental group than in the control group (Mayo 1991). The positive effect study of Mayo differed from the other studies, by being the only study which compared intensive NDT treatment (once a week) to less intensive NDT (once a month). The other eight studies compared NDT to infant stimulation (Palmer et al 1988, 1990) or to a not further defined form of standard care. In six of the seven studies motor outcome in the NDT group was similar to that of the contrast group. In the seventh study motor development was worse in children treated according to the principles of NDT than in children who received an infant stimulation programme (Palmer et al 1988, 1990). The above studies indicate that NDT during the first years of life does not have a measurable positive effect on motor development. This is in line with the conclusion of a recent review on the effects of NDT for people with cerebral palsy, aged 5 months to 22 years, that NDT did not have a clear beneficial effect on developmental outcome (Butler and Darrah 2001).

The other four high quality studies evaluated the effects of a developmental programme, treadmill training or conductive education. The two developmental programme studies (Barrera et al 1986, Leksculchai and Cole 2001) and the treadmill training study (Ulrich et al 2001) reported a positive effect of intervention on motor development. The fourth study compared the effect of conductive education with that of traditional neurodevelopmental programmes. Both types of intervention were associated with similar degrees of developmental progress (Reddihough et al 1998).

Treatment according to Vojta was only evaluated in two studies. D'Avignon and colleagues (1981) compared in a small randomised trial with a limited methodological quality the effect of treatment according to Vojta with NDT. They reported that the groups did not differ significantly in developmental outcome. Kanda et al (2004), who studied the effect of the amount of Vojta treatment on developmental outcome, reported a better outcome for the group which received sufficient Vojta training compared to the group which had had insufficient Vojta therapy. However, a major drawback of the study is that the design suffered from self-selection of the groups.

In conclusion, the current review indicates that intervention programmes in the first postnatal years according to the principles of NDT or Vojta do not have a beneficial effect on motor development in children at high risk for developmental disorders or children with CP or Down syndrome. However, substantial evidence has been provided that specific developmental training and general developmental programmes in which parents learn how to promote infant development can exert a positive effect on motor development.

Concluding Remarks

The present review indicates that intervention in children at risk of developmental disabilities should be adapted to the infant's age, *i.e.* the type of intervention which might be beneficial for infants at preterm age differs from the type which is effective in infants who have reached at least term age.

At preterm age infants seem to benefit most from intervention which aims at mimicking the intrauterine environment, such as the NIDCAP intervention. Some evidence has been provided that NIDCAP might have a temporary beneficial effect on infant motor (and cognitive) development. Future studies should address the question whether NIDCAP affects developmental outcome at school age.

The studies conducted after term indicated that intervention programmes according to the principles of NDT or Vojta, i.e., programmes in which passive handling techniques play a prominent role, do not have a clear beneficial effect on motor development. But intervention by means of specific motor training programmes, such as training of locomotor movements on a treadmill and general developmental programmes, where intervention aims at stimulation of the child's exploration of active motor behaviour, can exert a positive effect on motor development. Too few of these training and developmental programme studies were available to answer the question whether the age at which intervention starts matters, leaving this question for future research.

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CHAPTER 4

DOES EARLY INTERVENTION IN INFANTS AT HIGH RISK FOR A DEVELOPMENTAL MOTOR DISORDER IMPROVE MOTOR AND COGNITIVE DEVELOPMENT?

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Abstract

Infants at high risk for developmental motor disorders are in general referred to early intervention (EI) services. It is a matter of debate to which extent EI may facilitate outcome in various developmental domains. We reviewed the effects of EI programmes aiming at promoting motor and cognitive development. With respect to motor development the data indicated that EI prior to term age probably is most effective when it aims at mimicking the intrauterine environment; after term age general developmental programmes probably are most effective. Some evidence was provided that EI prior to term age has a beneficial effect on cognitive development regardless the type of intervention which is applied. After term age only general developmental programmes seemed to have an effect on cognitive development. The review concludes with preliminary data on the effect a new intervention programme, COPCA, applied between 3 and 6 months corrected age on developmental outcome till 18 months. The results indicated that COPCA was more beneficial for the development of sitting behaviour and cognition than traditional paediatric physiotherapy.

Keywords

Early intervention, motor development, cognitive development, developmental disorder, infant, COPCA

Introduction

Infants who are born with a low birth weight or preterm are at biological risk for developmental disorders (Aylward et al 1989, Bhutta et al 2002). Therefore these infants often are called high risk (HR) infants. In general HR infants have poorer cognitive, motor and behavioural outcomes than infants born at term. The less advantageous outcome is not only present at school-age, but persists into adolescence and adulthood (Bhutta et al 2002, Marlow 2004, Johnson et al 2005). The developmental outcome of infants born prematurely is heterogeneous and can be partly associated with biological risk factors, such as gestational age, birth weight, brain damage acquired in the pre- or perinatal period or subsequent illnesses. However, these factors account only for a portion of the variance associated with long term outcomes. In part the outcome is also mediated by environmental experience (Shonkoff and Meisels 2000, Vohr et al 2000, Aylward 2005, Johnson et al 2005, Spittle et al 2005). The complex character of influences on development makes it almost impossible to determine the relation between biological risk, environmental risk and neurodevelopmental outcome. Another factor which influences developmental outcome is the so-called plasticity of the brain. Depending on the timing of the insult to the brain and the size and site of the lesion functional recovery may occur. The brain is plastic in particular during the phase occurring after the completion of neuronal migration, during which the processes of dendritic outgrowth and synapse formation are highly active (Kolb et al 2001). The latter means that considerable plasticity can be expected between 2-3 months before until about 12-15 months after term age (de Graaf-Peters and Hadders-Algra 2006).

While major disorders such as mental retardation, epilepsy or cerebral palsy – which occur in 6-25% of HR infants (Aylward 2005) – are often identified during infancy, the effects of minor neurological dysfunction (MND) become more obvious with increasing age and are for instance expressed in learning disorders, attention deficit hyperactivity disorder (ADHD), clumsiness or other behavioural problems. MND occurs in as many as 50-70% of HR infants (Aylward 2005). Two basic forms of MND have been distinguished, i.e. simple and complex MND (Hadders-Algra 2002). Simple MND occurs relatively frequently and it is most probably caused by genetic predisposition and stress during pre- or perinatal life. Simple MND might be considered to represent the less optimal but still normal brain function. Complex MND is strongly related to perinatal adversities. It might be considered as a borderline form of CP and shows a clear association with motor, cognitive and behavioural problems (Hadders-Algra 2002, 2003).

The variety in developmental problems that may occur as a result of pre- and perinatal adversities makes it difficult to recognize infants at high risk for both major and minor neurological dysfunction at early age. The development of imaging techniques has improved the possibilities to identify these infants at early age (Aylward 2005). However, these techniques are not always able to visualize abnormalities in the brain (Hadders-Algra 2000b). Another instrument which might assist the early identification of HR infants is the assessment of the quality of General Movements (GMs) (Prechtl 2001, Hadders-Algra 2004). GMs are complex movement patterns involving head, trunk, arms and legs. They are present from early foetal life and disappear around 3-4 months

post term when goal-directed motor behaviour emerges. The quality of GMs, which is primarily determined by movement complexity and variation, reflects the integrity of the nervous system. Various studies indicate that the presence of definitely abnormal GMs around 3 months post-term, i.e. in the last phase of GM-development, is associated with a high risk for developmental motor disorders, such as CP (Prechtl et al 1997, Prechtl 2001, Hadders-Algra 2004, Groen et al 2005).

The early detection of infants at high risk for developmental disorders offers the opportunity for intervention at young age, i.e. during a phase in which the central nervous system is characterized by considerable plasticity. It is however still a matter of debate to what extent early intervention (EI) may facilitate outcome in various developmental domains. Therefore the first aim of the present paper is to review the effects of EI programmes which aim primarily at the improvement of motor development on a) motor and b) cognitive outcome. The analyses were restricted to interventions starting prior to 18 months corrected age. The third part of the paper summarizes the first results of an ongoing EI-study in which the effect of a recently developed physiotherapeutic intervention programme for infants with high risk for developmental disorders is compared to the effect of EI based on the principles of NeuroDevelopmental Treatment (NDT).

Effects of EI on motor development of HR infants

Recently we reviewed the literature on the effects of EI on motor development of HR infants (Blauw-Hospers and Hadders-Algra 2005). We first will summarize the findings of this review. This will be followed by a discussion and integration of additional studies which have subsequently been published. We started with a literature search which covered various databases, including Medline, CINAHL, PsycINFO and PEDro, in the period 1966 – June 2004. Thirty four studies were identified which fulfilled the following four criteria: a) subjects of study: infants with high biological risk for or with developmental disabilities, b) aim of the intervention (mostly amongst others): to improve motor development, c) onset of intervention in at least 50% of the participating children: between birth and the (corrected) age of 18 months, d) journal in which the study was published had an impact factor of > 0.3. Excluded were studies restricted to medical and orthopaedic interventions and studies in populations of healthy low-risk preterm infants or in populations of socially disadvantaged children without specific biological risk for developmental disorders.

The data revealed that a large diversity of EI programmes is applied. Before term age Newborn Individualized Developmental Care and Assessment Program (NIDCAP; Als et al 1994) is mostly used, after term age a whole range of specific and general developmental programmes are applied, together with more traditional physiotherapeutic interventions such as NeuroDevelopmental Treatment (NDT; Bobath 1980, Howle 2002) and treatment according to Vojta (Vojta 1976). Specific developmental programmes aim at the improvement of one aspect of motor development for example walking skills, while general developmental programmes aim at improvement in all developmental domains. Parents usually play an important role in the implementation of the specific and general developmental programmes.

The methodological quality (MQ) of each study was analysed systematically by taking into account 1) the level of evidence (Sackett 1989), 2) internal and 3) external validity. It was encouraging to note that 20 out of 34 studies had a high MQ, i.e. they had an evidence level I and a fair to high internal and external validity. This indicates a considerable improvement in the quality of studies evaluating the effect of early intervention (Siebes et al 2002). Previous studies indicated that with an increase in the rigorousness of the studies the evidence for the effectiveness of EI decreases (Parette and Hourcade 1984, Siebes et al 2002). For the studies included in the present review this also held true. Of the 20 studies with a high methodological quality only six (30%) showed a significant beneficial effect of intervention on motor development. From the 14 studies with a limited methodological quality, seven (50%) reported a positive effect of intervention.

The review indicated that intervention which might be beneficial for infants at preterm age is different from the type which is effective in infants who have reached at least term age. At preterm age infants seem to benefit most from intervention which aims at mimicking the intrauterine environment, such as NIDCAP intervention. After term age, intervention by means of specific or general developmental programmes has a positive effect on motor development. No evidence was found for a beneficial effect of traditional forms of paediatric physiotherapy like NDT and Vojta.

An additional literature search was performed to cover the period July 2004 – September 2006. The results of this search are shown in Table 4.1. Another five studies were identified which fulfilled the four aforementioned selection criteria. In two studies the onset of intervention was during the period of stay at the Neonatal Intensive Care Unit (NICU; Oghi et al 2004, Cameron et al 2005). Both studies had a fair methodological quality (MQ), consisting of a combination of level I evidence (Sackett 1989), a fair internal validity, and a limited external validity. The studies applied different intervention techniques in different groups of children and were unable to demonstrate a beneficial effect of intervention on motor development. The other three studies started between discharge from the NICU and the age of 9 months (Koldewijn et al 2005, Badr et al 2006, Gianni et al 2006). Gianni et al (2006) evaluated the effects of a mother-child intervention. The study had a fair methodological quality and was unable to demonstrate a beneficial effect of intervention on motor development. The other studies had a lower MQ. Badr et al (2006) studied the effects of a cognitive/sensorimotor stimulation programme on motor development, but was not able to demonstrate an effect of the intervention. The study of Koldewijn et al (2005) reported the results of a pilot study on the Infant Behavioral Assessment and Intervention Program (IBAIP). They demonstrated that at both 3 and 6 months motor development improved significantly compared to the control group. But due to the fact that they had used a historical control group the methodological quality of this study was limited (evidence level III, fair internal but low external validity).

We conclude that the five recent studies on the effect of early intervention in infants at high risk for developmental motor disorders do not alter the conclusions of the original review (Blauw-Hospers and Hadders-Algra 2005): intervention prior to term age probably is most effective when it aims at mimicking the intrauterine environment; intervention after term age probably is most effective when it consists of stimulation of motor development by means of the promotion of self-produced motor behaviour.

Table 4.1. Characteristics of additional studies (July 2004-September 2006) on the effect of EI on motor development.

Study characteristics

Study	*N*	*E*	*C*	*Attrition (%)*	*Study group*	*Evidence level*	*Internal validity*	*External validity*
NICU								
Ohgi et al. 2004	24	12	12	4	HR with CNS injury	I	+	-
Cameron et al. 2005	72	34	38	17	HR	I	++	+
After NICU – 9 mo								
Koldewijn et al. 2005	44	24	20	11	HR	III (historical control)	+	-
Gianni et al. 2006	36	18	18	0	HR	I	++	+
Badr et al. 2006	62	32	30	31	HR with CNS injury	I	+	-

Programme characteristics

Study	*Method*	*Descr.*	*Application period*	*Intensity*	*Location*	*Parents*
NICU						
Ohgi et al. 2004	EIP	++	prior to discharge - 6 mo	prior to discharge: 3-4x 30 min. After: 1x wk/ 2wk 40-60 min	Hospital/ Center	P↑
Cameron et al. 2005	Neonatal Dev pgm, largely based on NDT	++	birth - 4 mo	- hospital daily - depending on risk max 1x 2wk	Hospital / Home	P↑
After NICU – 9 mo						
Koldewijn et al. 2005	IBAIP	++	1 wk after discharge - 6 mo	3x evaluation, 6-8 home interventions	Home	P↑
Gianni et al. 2006	Mother-child intervention	+	3 mo - 12 mo	2x per month, 1.5 h	Center	P↑
Badr et al. 2006	Cognitive-sensorimotor stimulation program	+	discharge - 12 mo	1 mo: 2x wk, till 4 mo: 1x wk, thereafter 1x 2wk	Home	P↑

Evaluation moments, outcome measures and results

Study	*Ages of evaluation*					*Outcome measures*		*Results*[1]
	<Term	*Term*	*1-9 mo*	*9-18 mo*	*>18 mo*	*Neuromotor*	*Developmental*	
NICU								
Ohgi et al. 2004	1x		1,6			NBAS, Bayley PDI	Bayley MDI	E=C
Cameron et al. 2005			4			AIMS		E=C
After NICU – 9 mo								
Koldewijn et al. 2005		1x	3,6			NBAS, Bayley PDI, IBA	Bayley MDI	E>C
Gianni et al. 2006				12	24,36		Griffith	E=C
Badr et al. 2006			6	12, 18		Bayley PDI	Bayley MDI	E=C

C = Control group, CNS = Central Nervous System, E = Experimental group, EIP = Early Intervention Program, h = hour, HR = High risk, IBAIP = Infant Behavioral Assessment and Intervention Program, min = minutes, mo = months (corrected age), n = number, NDT = Neurodevelopmental Treatment, Neonatal Dev pgm = Neonatal Developmental program, P = parents, P↑= enhance parental skills, T = parents are 'therapist', wk = weeks, ? = no information available

Outcome measures: AIMS = Alberta Infant Motor Scale, Griffith = Griffiths Developmental Scales, IBA = ,MDI = Bayley Scales of Infant Development – Mental Development Index, NBAS = Neonatal Behavioral Assessment Score Bayley, Bayley PDI = Bayley Scales of Infant Development – Psychomotor Development Index,

[1] Statistically significant differences found in motor outcome at oldest age of evaluation: E > C: Experimental group significantly better outcome than Control Group, E = C: no difference between groups, E < C: Control Group better outcome than Experimental Group.

The table shows the characteristics of studies on early intervention focusing on motor development published between July 2004 and October 2006. In the table 'study characteristics' the sample size, study group and criteria for the methodological quality are presented. The second part 'programme characteristics' describes the methods of intervention together with the duration and intensity of treatment, the location where the treatment was applied and the role of the parents in the intervention programme. In the third part the ages at which developmental outcome is evaluated, the outcome measures to determine progress in motor development and the overall results of the EI programmes are shown (for detailed description see Table 3.3-3.5, Chapter 3, page 46-53.

Do EI programmes which focus on motor development affect cognitive outcome?

It is generally acknowledged that HR infants, as a group, have lower cognitive scores than typically developing infants and that their cognitive abilities may deteriorate over time (Vohr and Garcia Coll 1985, Aylward 1989, Bhutta et al 2002, Ment et al 2003). Therefore early intervention programmes often do not only aim at the improvement of motor outcome but also at the facilitation of cognitive development.

Over the years the effects of various general developmental intervention programmes aiming at the improvement of cognitive development have been studied extensively for both environmentally and biologically disadvantaged infants. Studies on early intervention by means of general developmental programmes in children who are environmentally disadvantaged indicate both a short and long term beneficial effect on cognitive development (Ramey and Campbell 1984, Wasik et al 1990, Campbell and Ramey 1994). The programmes designed for infants at biological risk for developmental disorders show less consistent effects of EI by means of general developmental programmes on cognitive development. For instance, the Vermont Intervention Program found both short and long term effects on cognitive function (Rauh et al 1988, Achenbach et al 1990, Achenbach et al 1993). Studies on the Infant Health and Development Program (IHDP) however indicated that immediately after the intervention at the age of three years a positive effect on cognitive development was present (Infant Health and Development Program 1990), but that the effect disappeared in the follow up at 5 and 18 years for the group with the higher biological risk status, i.e. the infants with a gestational age at birth of less than 37 weeks and a birth weight of less than 2000 grams (Brooks-Gunn et al 1994, McCormick et al 2006). In the children with the lower biological risk, i.e. who had a birth weight between 2001 and 2499 grams, the effect of intervention was still visible at the age of 18 years. The findings of the IHDP programme are consistent with the results of a more recent study on a general developmental programme, the Avon Premature Infant Project, which showed that the beneficial effect of the intervention which was observed at two years (Avon Premature Infant Project 1998) had disappeared when the study group was re-examined at five years of age (Johnson et al 2005). Thus general developmental programmes aiming at the improvement of cognitive development are indeed successful.

We wondered whether EI programmes focussing primarily on motor development have a similar effect on cognitive outcome. The hypothesis underlying this assumption is that improvement of early motor development allows the infant more opportunities to interact with the environment, which in turn may facilitate cognitive development (Becker 1999). To this end we re-examined whether the EI programmes which primarily aimed to improve motor development and were included in the review described in the previous section also evaluated cognitive outcome. To ensure the completeness of our study we carried out an additional literature search covering different databases including MEDLINE (1966 to November 2006), CINAHL (1982 to November 2006), AMED (1985 to November 2006), PsycINFO (1967 to November 2006) and PEDro. The keyword 'early intervention' was searched in combination with one of the following keywords: 'infant', 'motor development', 'cognitive development', 'low birth weight', 'preterm', 'high-risk', 'developmental disabilities' and 'cerebral palsy'. We used the same inclusion and exclusion criteria as in the review on motor development, be it that in the current review both motor and cognitive outcome measures had to be included. The search did not yield other studies than those already identified for the previous review. Twenty six studies of the previous 34 studies included in the review also had a cognitive outcome measure. The results of the effect of EI on cognitive development are summarized in Table 4.2.

In 13 of the 26 identified studies intervention had started in the NICU period, in eight studies intervention was initiated in the period between discharge from the NICU and the corrected age of 9 months and in three studies intervention started between 9 and 18 months. In two other studies the preset age criteria were not fulfilled i.e. they started at some age between discharge from the NICU and 18 months. All studies were reviewed on methodological quality (MQ). A fair MQ was present in sixteen studies. They all evaluated cognitive outcome with either the mental scale of the Bayley Scales of Infant Development (BSID) or with the Griffiths Developmental Scales.

Seven studies with a fair MQ evaluated outcome of intervention which had started during the NICU period. Five out of these seven studies reported a beneficial effect of intervention on cognitive development (Resnick et al 1987, 1988, Als et al 1994, Nelson et al 2001, Kleberg et al 2002, Tessier et al 2003). Interestingly the five studies evaluated four different types of early intervention programmes: Developmental Program including stimulation, NIDCAP, Auditory-Tactile-Visual-Vestibular Stimulation, and Kangaroo Care. The other six studies had a lower MQ. Three suggested a positive effect on cognitive development (Leib et al 1980, Als et al 1986, Feldman et al 2002). Six of the eight studies that started after discharge from the NICU but before the corrected age of nine months had a fair MQ. Two demonstrated that EI had a beneficial effect on cognitive outcome (Barrera et al 1986, Gianni et al 2006). Both studies evaluated the effects of a general developmental programme. The other four studies with fair MQ which did not find a positive effect of EI on cognitive development all addressed the effects of NeuroDevelopmental Treatment (Goodman et al 1985, Piper et al 1986, Rothberg et al 1991, Weindling et al 1996).

Table 4.2. Effect of early intervention on cognitive development.

Study	*Evidence Level*[1]	*Internal validity*[2]	*External validity*[3]	*Results motor*[4]	*Results cognitive*[4]
NICU					
Als et al. 1994	I	++	++	E > C	E > C
Resnick et al. 1987/1988	I	+	+	E > C	E > C
Ariagno et al. 1997	I	+	+	E = C	E = C
Charpak et al. 2001	I	+	+	E = C	E = C
Nelson et al. 2001	I	+	+	E = C	E > C
Kleberg et al. 2002	I	+	+	E = C	E > C
Tessier et al. 2003	I	+	+	E = C	E > C
Westrup et al. 2004	I	+	-	E = C	E = C
Ohgi et al 2004	I	+	-	E = C	E = C
Feldman et al. 2002	II	-	-	E > C	E > C
Als et al. 1986	III	+	-	E > C	E > C
Leib et al. 1980	III	-	-	E > C	E > C
Kleberg et al. 2000	III	-	-	E = C	E = C
After NICU – 9 mo					
Goodman et al. 1985	I	++	++	E = C	E = C
Piper et al. 1986	I	++	++	E = C	E = C
Barrera et al. 1986	I	++	+	E > C	E > C
Weindling et al. 1996	I	++	+	E = C	E = C
Rothberg et al. 1991	I	+	+	E = C	E = C
Gianni et al. 2006	I	++	+	E = C	E > C
Badr et al. 2006	I	+	-	E = C	E = C
Koldewijn et al. 2005	III	+	-	E > C	E > C
9 mo – 18 mo					
Palmer et al. 1988/1990	I	++	++	E < C	E = C
Eickmann et al. 2003	II	+	+	E > C	E > C
Piper and Pless 1980	III	+	-	E = C	E = C
After NICU – 18 mo					
Mayo 1991	I	++	+	E > C	E > C
Harris 1981	I	+	+	E = C	E = C

Table lay-out is based on Blauw-Hospers and Hadders-Algra 2005.

[1] Evidence level according to Sackett's method for grading research (Sackett 1989):
I = Randomised controlled trials; II = Non-randomised controlled trials / Prospective cohort studies with concurrent control group; III = Cohort studies with historical control group / Case studies with control participants

[2] Internal validity is determined by various study variations such as subject assignment, study design, attrition, confounders, validity of outcome measures, blind assessment

[3] External validity is determined by possibilities for generalization (group size and internal validity)

[4] Statistically significant differences found in motor outcome at oldest age of evaluation:
E > C: Experimental group significantly better outcome than control group
E = C: No difference between groups
E < C: Control group better outcome than experimental group

Only one of the three studies in which intervention started between 9 and 18 months had a fair MQ (Palmer et al 1988, 1990). This study demonstrated a beneficial effect of an infant stimulation programme on motor development, but the programme did not affect cognitive outcome. Both studies in which intervention started between discharge from the NICU and 18 months had a fair MQ. The study of Mayo (1991) on the effect of NDT demonstrated a significant improvement in cognitive function for the group which received the intensive treatment approach. The study of Harris (1981) addressed the effect of NDT in children with Down syndrome; it failed to demonstrate a positive effect on cognitive development.

In conclusion, the review provides some evidence that intervention during the NICU period in infants at high risk for developmental motor disorders has a beneficial effect on cognitive development. Interestingly, the effect seems to be independent of the type of intervention. The studies in which intervention had started after term age indicated that intervention by means of a general developmental programme might be beneficial for cognitive development. This finding is consistent with the results of studies on the effect of EI in infants with high biological risk for developmental disorders which primarily aim at improving cognitive outcome. Our review indicated that NDT applied during infancy does not produce benefits in the cognitive domain – a finding in line with an earlier review of Butler and Darrah (2001).

The early intervention project in Groningen (VIP project)

The review above indicates that convincing evidence for a beneficial effect of physiotherapy programmes most frequently used in early intervention (NDT and treatment according to Vojta) is lacking. This inspired two members of the Groningen team (TD and MHA) to develop a new physiotherapeutic EI programme: COPCA (Coping with and Caring for infants with neurological dysfunction – a family centred programme). COPCA is based on the principles of the Neuronal Group Selection Theory (NGST; Edelman 1993) and on new insights in the field of education and family care (Dale 1996, Rosenbaum et al 1998). According to NGST typical development is characterized by two phases of variability, i.e. primary and secondary variability. During primary vari-

ability the nervous system explores all motor possibilities available. In typically developing infants this phase is characterized by abundant variation. At function specific ages the infant reaches the phase of secondary variability. The child gradually learns to select the most efficient solution for a given task out of his motor repertoire. This selection is based on trial and error. This means that during the phase of secondary variability the child learns to adapt his/her motor behaviour to specific situations (Hadders-Algra 2000a). Infants with a brain lesion resulting in a developmental motor disorder have a reduced repertoire of motor strategies available for exploration. In addition, these infants have problems with the selection of the most appropriate solution for a certain task out of the repertoire (Hadders-Algra 2000a). Therefore COPCA aims to promote variation in motor behaviour and trial-and-error experiences, with the ultimate goal that the child will be able to find an appropriate solution for any motor task.

An equally important base of COPCA are new insights in the field of education and family care (Dale 1996, Law et al 1998, Rosenbaum et al 1998). Family centred care implies a partnership between the family and the professional, in which the family defines the priorities for intervention, while the therapist assists the family throughout the intervention period (Law et al 1998). In COPCA the physiotherapist (PT) acts as a coach who respects the autonomy of the family and intends to make him- or herself superfluous. Caregivers are key-persons in the COPCA programme. They determine the goals to be achieved. The PT provides information on infant development, the specific problems of infants with neurological dysfunction, basic principles on infant education, and specific ways in which development may be promoted and problems may be tackled. By means of a continuous dialogue with the PT, caregivers thus develop their own ways to cope with their infant.

In the so-called VIP project (VIP is the abbreviation of the Dutch term for Early Intervention Project, namely Vroegtijdige Interventie Project) the effect of COPCA on developmental outcome till the age of 18 months is currently studied. Here we will present preliminary data on the first 20 infants included in the project who have already reached the age of 18 months. The aim of the study is to assess whether COPCA is more beneficial for developmental outcome at 18 months corrected age than traditional paediatric physiotherapy (TPP), which in the Netherlands in general consists of the implementation of the 'living concept' of NDT. The latter implies that TPP is practised with significant heterogeneity.

The study groups of the VIP project consisted of infants who had been admitted to the NICU of the University Medical Center Groningen (UMCG) between March 2003 and May 2005 and showed at ten weeks corrected age definitely abnormal GMs. As indicated in the introduction the presence of definitely abnormal GMs indicates a high risk for developmental motor disabilities, such as cerebral palsy (CP) (Prechtl 2001, Hadders-Algra 2004). Infants with severe congenital anomalies and infants whose parents had an inappropriate understanding of the Dutch language were excluded from the study. All parents of the infants gave informed consent and the project was approved by the Ethics Committee of the University Medical Center Groningen.

The infants were randomly assigned to the COPCA or the control group. The randomised intervention period lasted from 3 to 6 months corrected age[1]. Thereafter the intervention followed the suggestions of the paediatrician in charge of the care of the infant. COPCA intervention was provided twice a week for one hour in the home environment by one of four specially trained paediatric physiotherapists. The control group received TPP with a frequency varying from 2 to 20 times (median value: 7) during the intervention period. The duration of the TPP sessions varied from 15 to 36 minutes (median value: 30); in general they were also applied in the home environment. During the study the assessors were blind regarding group allocation.

Of the first twenty infants enrolled in the VIP project nine had been assigned to COPCA intervention and 11 to the control group. The latter got physiotherapeutic guidance indicated by the paediatrician. It turned out that nine infants in the control group had received TPP and two got no specific guidance. As the results of the two infants who did not receive TPP did not differ from those who did, we decided to include these two infants without TPP in the control group. The COPCA and control group were similar in terms of gender distribution, gestational age at birth and birth weight (Table 4.3). All but two infants had been born preterm.

Table 4.3. Clinical characteristics of the two study groups.

	COPCA (n = 9)	TPP (n = 11)
Gender	4 male	5 male
	5 female	6 female
Preterm (GA<37 wk)	n = 8	n = 10
Term	n = 1	n = 1
Gestational age (wk)	27-37	27-39
	(median 29)	(median 31)
Birth weight (g)	585-3640	825-3460
	(median 1160)	(median 1340)

[1] Throughout the rest of the document ages in months are ages corrected for preterm birth.

The infants' developmental status was assessed with a battery of tests at 3 months (baseline), 4, 5, 6 months and 18 months. At each age the assessment included a standardized neurological examination according to Prechtl (1977) with age-specific adaptations of the norms according to Touwen (1976) or at 18 months according to Hempel (1993) and the Alberta Infant Motor Scale (AIMS; Piper and Darrah 1994). The neurological findings were summarized as normal, minor neurological dysfunction (MND) or abnormal. During early infancy abnormal indicated the presence of a full-blown neurological syndrome, such as a clear hypo- or hypertonia, a hemisyndrome, or a hyperexcitability syndrome; at 18 months it denoted the presence of CP.

MND indicated the presence of signs of neurological dysfunction not meeting the criteria for clear neurological pathology. The signs of MND were grouped into the following functional clusters: mild abnormalities in gross motor function, mild abnormalities in fine motor function, mild abnormalities in muscle tone regulation, mildly abnormal reflexes, and mild cranial nerve dysfunction (Hadders-Algra 2003). Age specific criteria for deviancy in a specific cluster were determined. On the basis of the number of clusters meeting the criteria for deviancy two forms of MND could be distinguished: simple MND, which denoted the presence of one cluster of dysfunction and complex MND, when more than one cluster fulfilled the criteria for deviancy (Hadders-Algra 2002, 2003). At the age of 18 months we also applied the optimality concept to summarize neurological condition (Huisman et al 1995). For 57 items, which represent the neurological examination, criteria for optimality were defined. It is important to realize that the definition of optimal is narrower than that of normal or typical. The Neurological Optimality Score (NOS) of an infant is the sum of items meeting the criteria for optimality, which can count up to a maximum of 57 points.

The AIMS (Piper and Darrah 1994) was used to evaluate gross motor development in various positions (supine, prone, sitting and standing) over time. The 58 items each describe three aspects of motor performance – weight-bearing, posture and antigravity movements. The assessment results in a total score and subscores for the various positions. At 6 and 18 months also the Mental Scale of Bayley Scales of Infant Development (BSID-II; Bayley 1993, Van der Meulen et al 2002) was administered. The mental scale consists of items concerning discrimination, classification, language, social skills, memory and problem solving. The raw scores of the mental scale of the Bayley were converted into age-equivalent scores, as derived from the Dutch norms (Van der Meulen et al 2002).

The results showed that both groups did not differ in neurological condition. This was true for baseline, for short term outcome at 6 months and for long term outcome at 18 months. At 18 months, two children in each group were diagnosed with CP. All other infants showed the complex form of MND (Figure 4.1). As an important part of the neurological examination consists of the assessment of muscle tone, reflexes and cranial nerve function, it could perhaps be that both types of intervention have a similar effect on these neural functions. Therefore, we also analyzed neurological condition at 18 months by means of the neurological optimality score. The neurological optimality score focuses on the quality of motor behaviour in terms of variability and fluency.

The analysis of the optimality scores was applied only in the children without CP. The results suggested a small, but statistically not significant advantage for the infants of the COPCA group (Figure 4.2).

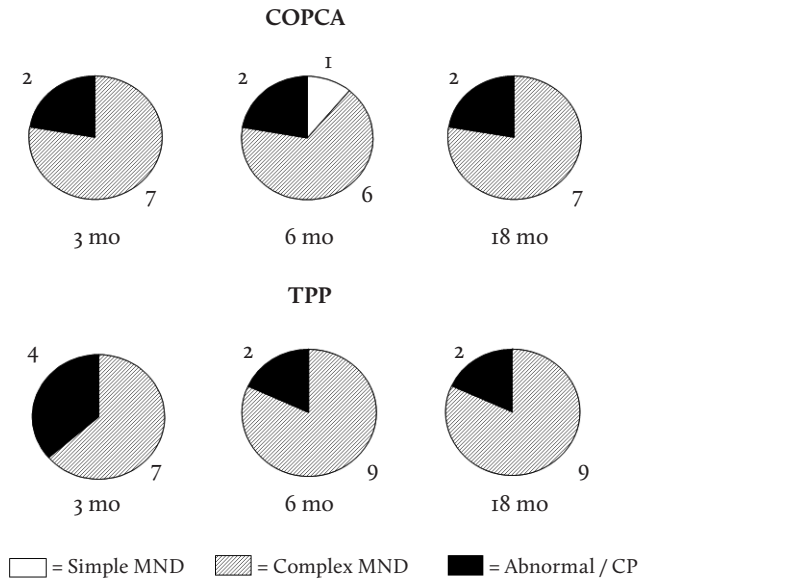

Figure 4.1. Neurological condition of infants of the COPCA and TPP group at 3, 6 and 18 months. The numbers refer to the actual number of infants.

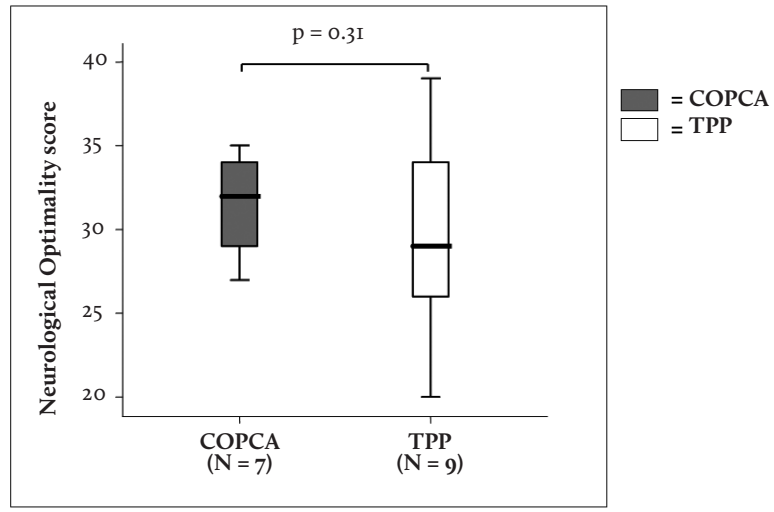

Figure 4.2. Effect of type of intervention on the neurological optimality score at 18 months in children without cerebral palsy. Vertical bars, range; horizontal lines, median value; boxes: interquartile range. Mann-Whitney U test: p=0.31.

The effect of intervention on the development of gross motor skills was analyzed with the AIMS. The data revealed that at the age of 18 months the AIMS suffers from a ceiling effect (Darrah et al 1998, Liao and Campbell 2004). We therefore restricted our analyses to the data of 3 to 6 months. The analyses were carried out twice, once for all infants and once without the children who developed CP. The rationale behind the second series of analyses was that it is conceivable that children with mild to moderate dysfunction profit more from intervention than children with severe dysfunction. The analyses showed no differences in total AIMS scores between the COPCA and the control group at baseline, during and immediately after the intervention period (Figure 4.3). Neither did the groups differ in scores on the subscales which assess motor behaviour in supine, prone and standing position. On the sitting subscale however a significant positive effect of the COPCA intervention was observed whether or not the infants who developed CP were included into the analysis (p=0.03 with CP included and p=0.02 with CP excluded; Figure 4.4a, 4.4b). Furthermore, the effect of COPCA intervention tended to increase with increasing age when children with CP were left out of the analysis (p=0.08) (Figure 4.4b). The results suggest that the infants who received COPCA intervention have a better postural control at 6 months than the infants of the TPP group. These findings match with the results of de Graaf-Peters et al (2006), who demonstrated by means of EMG recordings that the infants who received COPCA intervention showed at 6 months a more typical pattern of postural control during sitting than the infants of the TPP group.

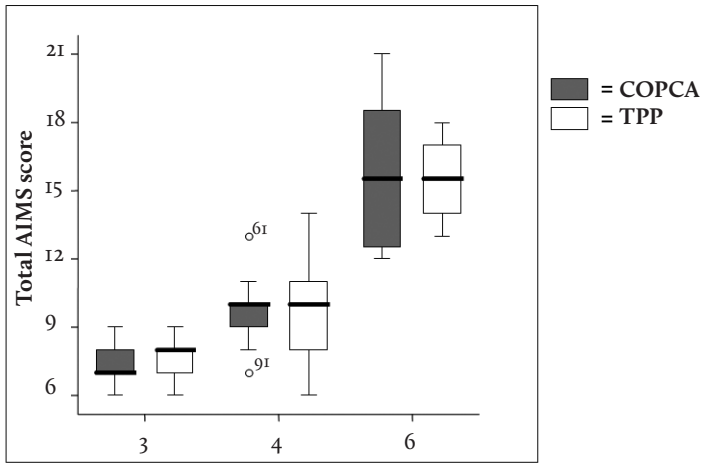

Figure 4.3. Effect of type of intervention on the total AIMS score at 3, 4 and 6 months. Vertical bars, range; horizontal lines, median value; boxes: interquartile range.

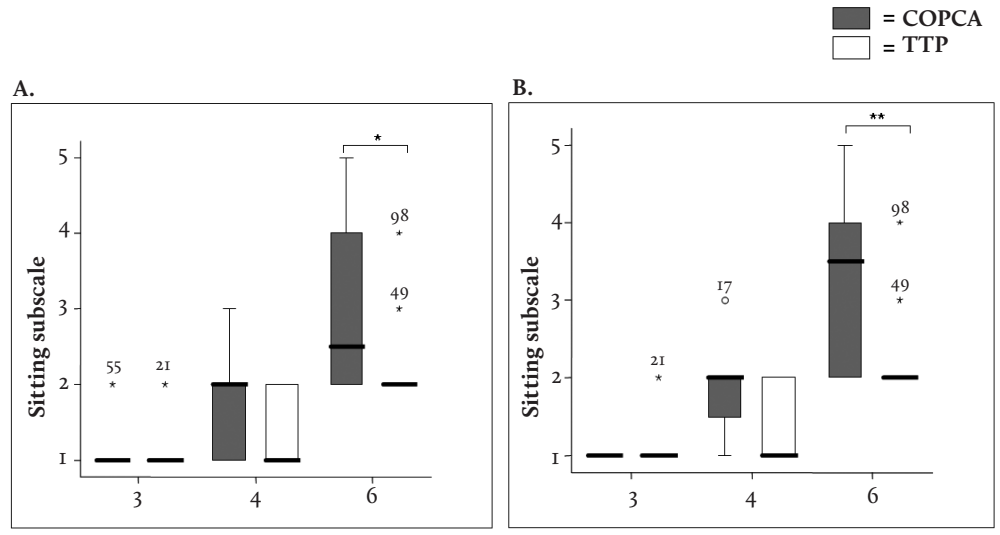

Figure 4.4 Effect of type of intervention on the sitting subscale of the AIMS at 3, 4, and 6 mo: a) with infants with CP included, b) with infants with CP excluded. Vertical bars, range; horizontal lines, median value; boxes: interquartile range. MANOVA: main effect of intervention * p=0.03, ** p=0.02. Intervention and age tended to interact (p=0.08).

Finally, the results showed that COPCA intervention might also have an effect on cognitive development. At 6 months the MDI of both groups was identical. In the TPP group we found a significant decrease in the median MDI score over time from 109 at 6 months to 99 at 18 months (p=0.03). In general it is known that cognitive abilities of preterm infants deteriorate with increasing age (Vohr and Garcia Coll 1985, Aylward 1989, Bhutta et al 2002, Ment et al 2003). In contrast to the TPP group, the MDI score of the infants who had received COPCA intervention remained stable over time (median values at 6 months 110, at 18 months 111; Figure 4.5). This indicates that COPCA intervention might be able to counteract a cognitive deterioration.

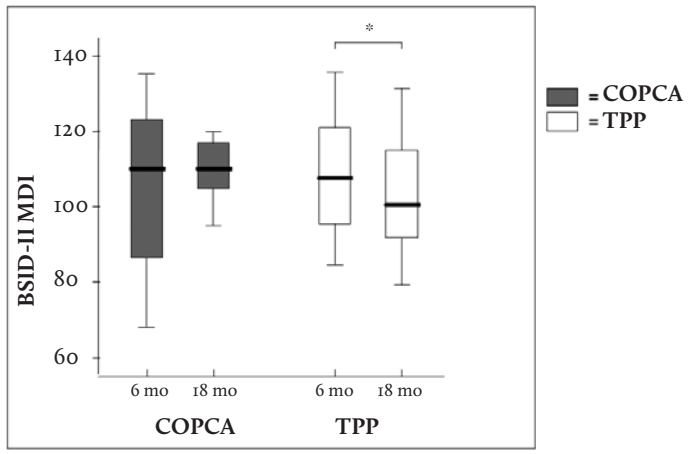

Figure 4.5 Bayley Scales of Infant Development: MDI scores at 6 and 18 months in COPCA and TPP group. Vertical bars, range; horizontal lines, median value; boxes: interquartile range. Wilcoxon: * p=0.03.

Concluding remarks

Prior to term age intervention which aims at mimicking the intrauterine environment is probably most effect for motor development; for cognitive development any type of intervention seems to be beneficial. After term age EI probably is most effective for motor and cognitive development when a general developmental programme is applied. No evidence is available supporting the notion that intervention according to the principles of NDT has a positive effect on motor or cognitive development. The limited amount of evidence favouring EI encouraged two members of the Groningen team (TD and MHA) to develop a new physiotherapeutic intervention, COPCA. Preliminary results indicate that COPCA has a significant effect on the development of sitting abilities and that COPCA might be able to counteract a cognitive deterioration. However, further research is needed to explore clinical effectiveness of the COPCA programme.

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CHAPTER 5

DEVELOPMENT OF A QUANTITATIVE TOOL TO ASSESS THE CONTENT OF PHYSICAL THERAPY FOR INFANTS

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Abstract

Purpose. The study aim was to describe and quantify physical therapy interventions for infants at high risk for developmental disorders.

Methods. An observation protocol was developed based on knowledge about infant physical therapy and analysis of directly observable physiotherapeutic (PT) actions. The protocol's psychometric quality was assessed. Videos of 42 infant physical therapy sessions at 4 or 6 months corrected age were analyzed.

Results. The observation protocol classified PT actions into 8 mutually exclusive categories. Virtually all PT actions during treatment could be classified. Interrater and intrarater agreement were satisfactory (intraclass correlations, 0.68-1.00). Approximately 40% of treatment time was spent on challenging the infants to produce motor behavior by themselves whereas approximately 30% of time facilitation techniques were applied. Tradition-based sessions could be differentiated from function-oriented ones.

Conclusions. It is possible to document PT actions during physical therapy treatment of infants at high risk for cerebral palsy in a systematic, standardized and reliable way.

Key Words

Physical Therapy, Infants, Cerebral Palsy, Program evaluation, Video observation, Quantitative tool, Reliability

Introduction and purpose

In pediatric physical therapy, multiple intervention programs are used for the treatment of infants with or at risk for developmental disorders, such as cerebral palsy (CP). Programs frequently used during a stay in the neonatal intensive care unit are the Newborn Individualized Developmental Care and Assessment Program, Kangaroo Care and sensory stimulation programs. Programs used after term age are quite different in approach and vary from treatment according to Vojta, to NeuroDevelopmental Treatment (NDT), to multiple forms of specific and general developmental programs (Blauw-Hospers and Hadders-Algra 2005). Four recent reviews (Butler and Darrah 2001, Blauw-Hospers and Hadders-Algra 2005, Blauw-Hospers et al 2007, Spittle et al 2007) did not find conclusive evidence that early physical therapy intervention improves motor outcome in children with or at risk for CP. Literature also does not provide evidence that early physical therapy produces other benefits, such as the enhancement of social-emotional or cognitive function (Butler and Darrah 2001, Blauw-Hospers et al 2007).

A "theory driven evaluation" (Chen 1990) might help to understand why evidence for the effectiveness of early intervention (EI) programs is lacking. Theory driven evaluations pay attention to the mechanisms underlying outcome. This means that they do not only evaluate the effect of intervention on outcome (eg, "does application of EI result in a higher score on the Bayley Scales of Infant Development?"), but also the pathway along which the outcome is achieved (eg, "is application of a specific physiotherapeutic (PT) action related to outcome?").

Theory driven evaluation offers a framework to understand the relation between program inputs, the contents of the program - the 'black box' -, and the outcomes (Brazil et al 2005). Knowledge about the effect of specific elements of an intervention is crucial for improvement of physical therapeutic guidance. Guidance should focus on elements which have a beneficial effect on outcome and elements which do not affect outcome or have an adverse effect should be discarded. Uncovering the actual contents of practiced physical therapy will shed light on potentially effective and ineffective elements of the intervention.

The aim of the present study is to develop an instrument to objectify what physical therapists actually do during treatment, that is, to open the black box of physical therapy sessions in daily practice, applied to infants at high risk for a developmental disorder such as CP. It should be noted that CP develops in only a portion of infants at high risk for developmental disorders. Yet, motor problems and difficulties in learning develop in a substantial proportion of these infants at school age (Aylward 2005).

To this end, we developed an observation protocol based on the theoretical constructs of the most commonly used physical therapy approaches for high risk infants in the Netherlands, that is, NDT and functional therapy (Ketelaar et al 2001, Ekström et al 2005). The term *functional* in the latter approach may give the impression that NDT does not aim at improving the infant's function. This is a false impression; all approaches in infant physical therapy aim at improving the child's function. Nevertheless, some differences exist between NDT and functional therapy. For instance, NDT is characterized by a broad repertoire of therapist-infant contact strategies,

whereas the functional approach relies entirely on infant self-produced motor behavior. Primary questions addressed in this study deal with the psychometric properties of the protocol. 1) Is the observation protocol complete, that is, is it possible to classify the majority (> 90%) of the contents of physical therapy sessions? 2) Are the observation items mutually exclusive? 3) Can the items be assessed reliably in terms of inter- and intraassessor agreement? Secondary questions address the actual content of the physical therapy sessions during early infancy: 4) Which observable therapeutic strategies can be distinguished? 5) What proportion of time is spent on the different strategies?

Methods

Development of the observation protocol

Protocol development started with a systematic analysis of the literature on the application of physical therapy in infants with or at risk for developmental motor disorders such as CP. This analysis gave us clues to the most important strategies that are used by pediatric physical therapists. These strategies are based on the theoretical constructs of the most commonly used physical therapy programs for high-risk infants, that is, NDT and functional therapy. The NDT approach was developed in the 1940s by Karl and Berta Bobath. Inhibition of spasticity and facilitation of normal posture and motor behavior were the main aims of treatment, which the Bobaths tried to accomplish by tone-influencing patterns with the use of various therapeutic handling techniques (Bobath and Bobath 1984, Mayston 2001). Over the years, NDT gradually evolved based on new theoretical constructs and practical knowledge. Although the basic philosophy and the approach to treatment remained the same, goals, treatment techniques and treatment application obtained a more functional orientation (Mayston 2001, Howle 2002). Currently the main goals of NDT are to increase or improve the skill repertoire, to maintain the functional level of performance, and the general management and minimization of contractures and deformities (Mayston 2001).

The way in which NDT is applied depends on the child's age and the severity of the disability. Already in the 1960s Elizabeth Köng and Mary Quinton started to adapt and develop techniques for early treatment of babies based on the Bobath concept (Bly 1999, Quinton and Nelson 2002). The principles of NDT baby treatment (Bly 1999) aim at helping the infant's motor development by giving the infant sensorimotor experiences of typical motor patterns. This is achieved by using several forms of facilitation techniques, such as handling or pressure techniques, by using support devices or by giving sensory experience to the infant. Therapy should not be restricted to the therapy sessions; the therapist aims by means of training the caregivers to achieve a carry-over from treatment into activities of daily life (Bobath and Bobath 1984, Finnie 1996, Mayston 2001).

The focus of functional therapy lies on improvement of functional tasks (Ketelaar et al 2001, Ekström et al 2005). Application of the concept of functional therapy to young infants implies that the cornerstone of guidance consists of encouragement of self produced motor behavior during functional tasks, such as feeding, dressing, bathing and playing (Resnick et al 1987, Palmer et al 1988, Ketelaar et al 2001, Ekström et al 2005). From the literature, it is known that infants at high risk for a developmental motor disorder have a reduced repertoire of motor strategies available for exploration. This is already expressed during the first postnatal months in a limited repertoire of general movements (GMs) and continues when goal-directed motility emerges. The infants also have difficulties in selecting the most appropriate solution for a certain task out of their motor repertoire (Hadders-Algra 2000b). Functional therapy therefore aims through play to promote variation in motor behavior and opportunities to explore the motor possibilities to find appropriate solutions for motor tasks (Hadders-Algra 2000b, Ketelaar et al 2001, Damiano 2006). Part and parcel of the more functional approach is the coaching role of the therapist, that is, the caregivers determine how developmental strategies highlighted by the therapist may be integrated into the child and family's daily life.

In the next stage of protocol development we analyzed 20 pilot video recordings of infant treatment sessions in terms of directly observable PT actions. All actions that physical therapists performed were documented. These PT actions were classified and defined into categories. The categories varied from classic NDT actions such as handling techniques to categories describing functional activities (self produced motor behavior) and family involvement and education. The observation protocol is the final result of a cyclical process that incorporated knowledge about physical therapy for infants and the observation of overt and directly observable therapeutic actions during treatment from video recordings. During the development of the observation protocol, we paid attention to required psychometric requirements: completeness, mutual exclusiveness, reliability, and construct validity (Reynders 1992).

Subjects

The contents of physical therapy sessions were analyzed for a study group of 22 infants, who had been admitted to the neonatal intensive care unit of the University Medical Center Groningen. All infants had an indication for early physiotherapeutic intervention on the basis of the presence of definitely abnormal GMs at the age of ten weeks corrected age. The GM method is a standardized technique to assess neurological integrity on the basis of the quality of spontaneous motor behavior (Einspieler et al 2004, Hadders-Algra 2004). GMs are spontaneously generated complex movements involving the head, trunk, arms, and legs. Four classes of GM quality can be distinguished: normal-optimal, normal-suboptimal, mildly abnormal and definitely abnormal. Various studies showed that the presence of definitely abnormal GMs at 2 to 4 months post-term indicates a high risk for developmental disabilities, such as CP (Prechtl et al 1997, Prechtl 2001, Hadders-Algra 2004). The GM assessment has a good predictive validity for both major and minor developmental disorders. Also construct and concurrent validity and reliability of the GM method are satisfactory (Heineman and Hadders-Algra 2008). Infants with severe congenital anomalies and infants whose caregivers had an inappropriate understanding of the Dutch language were excluded from the study. All caregivers of the infants signed an informed consent and the research project was approved by the Ethics Committee of the University Medical Center Groningen.

Twenty of the infants were born preterm with a gestational age at birth that varied from

28 to 32 weeks (median, 31) and a birth weight between 630 and 2090 g (median, 1205 g). Two infants were born at term with a median birth weight of 3560 g. Neurological condition was assessed at 6 months corrected age (CA) with the Touwen Infant Neurological Examination (TINE) (Hadders-Algra et al 2009). Three infants showed clear neurological dysfunction. Fourteen infants showed a high number of signs of minor neurological dysfunction (MND). The remaining five infants had a normal-suboptimal neurological condition at 6 months (Table 5.1).

Table 5.1. Birth characteristics and neurological outcome (n=22).

	Total (n = 22)	
Gender	10 boys	
	12 girls	
Gestational age (wk)	Preterm (n=20)	Term (n = 2)
Median	31	38
Range	28-32	37-39
Birth weight (g)	Preterm (n=20)	Term (n =2)
Median	1205	3560
Range	630-2090 g	3460-3660 g
Neurological examination at 6 mo*	4 Normal-suboptimal	
	15 MND	
	3 Abnormal	

* corrected for prematurity

g = gram, MND = Minor Neurological Dysfunction, mo = months, wk = weeks

Recording of intervention sessions

All 22 infants received physical therapy between the ages of 3 to 6 months corrected age. Therapy was delivered by 17 physical therapists who were registered in the Netherlands as pediatric physical therapists. Fifteen therapists treated the infants in their home environment and 2 applied the intervention in a clinical setting. Treatment duration varied from 12 to 50 minutes per session (mean, 30 minutes per session; SD 9 minutes per session).

At 4 and 6 months corrected age, a video recording of a treatment session of each infant was made. To minimize intrusion, the camera was positioned as far away from the physical therapist and infant as possible. In addition, therapist and caregiver were asked to ignore the person making the video so that the recording would resemble a natural situation. Research has shown that video recording hardly affects the behavior of people that are being filmed because they tend to forget the presence of the camera after awhile (Albrecht et al 2005). Care was taken that physical therapist, caregiver, and infant were continuously in view of the camera.

The video recordings were analyzed with the standardized observation protocol with help of the Noldus software program The Observer (Version 5.0; Noldus, Wageningen, the Netherlands), a program specially designed for behavioral observation. The program allows the quantification of the duration, frequency and serial order of defined therapeutic actions. For example, imagine the following sequence of actions: A) a physical therapist offers an infant a toy in the midline; B) after 6 seconds the therapist facilitates rolling behavior with the pelvis or legs as keypoint, C) meanwhile the therapist gives the caregiver instructions on handling during rolling movements. In the Observer action A is scored as category H (see Appendix 1, p. 188; the infant is challenged to produce motor behavior by himself or herself; activity flows over into therapeutic handling). As the toy is offered in the midline only, the action is performed with little variation (H1). Action B is scored as category C1 (facilitation techniques - handling), but at the same time the physical therapist interferes with the infant's behavior and interrupts his or her activity (category A.4.1). Action C is scored as category A.4.3 (caregiver training). Start and stop of an action are indicated by pressing a key on the computer keyboard. It is important to note that The Observer program allows scoring of multiple actions occurring simultaneously (eg, action B). To compare the results of different treatment sessions, the duration of the PT actions was converted into a relative duration, which represented a percentage of the total treatment time. Relative duration (%) = (Time spent on PT action / Total time of treatment session) * 100.

Psychometric quality of the observation protocol

Completeness of the protocol was tested by checking whether all possible PT actions observed by the researchers were covered by the categories of the observation protocol. Completeness was considered satisfactory if major part of the PT actions (> 90% of the observation time) could be classified into protocol categories. Mutual exclusiveness was determined by means of the inter- and intrarater agreement. Substantial agreement indicates that a single PT action is clearly related to a specific category of the observation protocol. Reliability of the observation protocol

was measured through inter- and intrarater agreement. Three time intervals, each lasting five minutes, were selected from within each of 5 randomly selected treatment sessions (n=15). The intervals 100-400 seconds, 500-800 seconds and 1000-1300 seconds were analyzed. The start of the treatment session, that is, the first 100 seconds, was excluded from the analyses because during this period mainly preparatory actions were performed. Interrater agreement was assessed by comparing observational scores of the third author with those of the first author. Intrarater agreement was assessed by comparing the observational scores of the first author who reanalyzed the video sequences with an interval of at least three weeks.

Data Analysis

The data were analyzed using the computer package SPSS (version 14.0; SPSS Inc, Chicago, Illinois). Inter- and intrarater agreements were calculated by intraclass correlations (ICCs) of frequency and relative duration of PT actions. ICC values between 0.50 to 0.75 were considered to indicate satisfactory reliability, ICC values exceeding 0.75 indicate good reliability (Portney and Watkins 2000).

For the comparison of the relative duration of the main categories and subcategories of PT actions and the amount of postural support at 4 months with the data at 6 months, Wilcoxon signed rank tests were used.

To get an impression of the current state of the daily practice in pediatric physical therapy in The Netherlands we analyzed whether sessions were dominated by PT actions that are in line with the concept of NDT in baby treatment[13] or by PT actions belonging to the functional approach. A treatment session was classified as NDT-like when the time spent on the category facilitation techniques exceeded the 75th percentile of current group data. Similarly, sessions in which time spent on the category 'challenging the infant to produce motor behavior by himself or herself - action continued by the infant' exceeded the 75th percentile were classified as functional sessions.

Correlations and differences with a p-value < 0.05 were considered as statistically significant.

Results

Observation protocol

The observation protocol contains three levels of observation. The first level consists of eight mutually exclusive main categories of PT actions (see Appendix 1). The second level consists of the subcategories of the main techniques. For example, the main category facilitation techniques has the following subcategories: handling, pressure techniques, transitions and support devices. The third level of the protocol entails concrete PT-actions. For example, in the supine or sitting position: shoulders function as a key point, handling hands guide the shoulders of the infant in protraction to control the infants movements and to facilitate hand-hand contact and symmetry. The degree of postural support provided by the physical therapist or caregiver when the infant was in prone, side or sitting position was scored as an additional variable. Postural support was classified into four categories: no postural support, minimal postural support, clear postural support and full postural support

Psychometric properties

The contents of 42 physical therapy sessions were analyzed. For two infants, only one video recording was available for observation. In one case, the therapist had already discontinued treatment before 6 months. In the other case, the family was on holiday when the video recording was scheduled.

Major part of the observed PT actions could be classified into the categories of the observation protocol. Only 3% of the duration of physical therapy sessions could not be classified into the categories of the observation protocol, that is, they were classified in the category 'not specified'. This indicates that the observation protocol meets the criterion for completeness.

Inter- and intrarater agreement on the frequency of PT actions was moderate to good with ICCs varying from 0.68 to 1.00 (interrater) and 0.70 to 0.98 (intrarater). The same was true for the inter- and intrarater agreement on the relative duration of actions; ICCs ranged from 0.76 to 1.00 (interrater) and 0.69 to 0.99 (intrarater; Table 5.2). The confidence intervals of the PT actions are presented in Table 5.2. The data indicate that the PT actions of the observation protocol can be assessed in a reliable way. Moreover the moderate to good inter- and intrarater agreement points to a sufficient degree of mutually exclusiveness.

Relative duration of therapeutic actions

The data on frequency and relative duration of PT actions were highly correlated (Spearman rank correlation at 4 months: 0.89, at 6 months: 0.88). This suggests that both parameters carry virtually identical information. We therefore decided to restrict our report on the contents of the sessions to one parameter. We chose relative duration as it had slightly better inter- and intrarater agreement values than frequency.

The relative duration of the majority of the PT actions applied at 4 months did not differ significantly from those at 6 months (Table 5.3). The only exceptions were challenging the infant to produce motor behavior by himself which flows over into handling, which occurred slightly more at 6 months than at 4 months (p=0.03) and in the provision of postural support. Less postural support was provided during PT actions in infants aged 6 months than in those aged 4 months (Table 5.3).

In the following report on the contents of the NDT sessions, we pooled the 4- and 6-month data for those actions in which the relative duration of PT actions at 4 and 6 months were identical. Table 5.3 shows that the application of physical therapy in infancy is characterized by a large heterogeneity. Most of treatment time was spent with PT actions in the categories of motor behavior produced by the infant, that is, motor behavior that was initiated by the infant and behavior that was challenged by caregiver or physical therapist who a) let the infant try on her or his own or b) took over control of the infant's movements by means of handling (together ~40%), and facilitation techniques (29%; Figure 5.1). We noted that during both challenging the infant to produce motor behavior by himself/herself, which flows over into handling (5.6%) and challenging the infant to produce motor behavior by himself/herself which is continued by the infant (13.2%),

Table 5.2. Interrater and intrarater agreement (ICC) on main and subcategories of physiotherapeutic actions (frequency and relative duration of PT actions; n=15 video fragments of 5 minutes).

Intervention	Interrater agreement		Intrarater agreement	
	Frequency (n=15)	Duration (n=15)	Frequency (n=15)	Duration (n=15)
	ICC (95% CI)	ICC (95% CI)	ICC (95% CI)	ICC (95% CI)
Family involvement and Educational component				
Educational actions	0.78 (0.48-0.92)	0.78 (0.48-0.92) [b]	0.80 (0.49-0.93) [b]	0.88 (0.66-0.96) [b]
- Caregiver interferes with infant's activities	n.o. [a]	n.o. [a]	n.o. [a]	n.o. [a]
- PT interferes with infant's activities	0.76 (0.43-0.91)	0.76 (0.43-0.91) [b]	0.89 (0.70-0.96) [b]	0.80 (0.51-0.93) [b]
- PT guides the infant	1.00 (1.00-1.00)	0.99 (0.98-1.00) [b]	0.74 (0.38-0.90) [b]	0.75 (0.40-0.90) [b]
- PT gives caregiver training	n.o. [a]	n.o [a]	0.95 (0.86-0.98) [b]	0.78 (0.46-0.92) [b]
- PT coaches the caregiver	n.o. [a]	n.o. [a]	n.o. [a]	n.o. [a]
Communication	0.87 (0.66-0.95)	0.98 (0.94-0.99) [b]	0.89 (0.69-0.96) [b]	0.96 (0.80-0.98) [b]
- Information exchange	0.87 (0.68-0.95)	0.98 (0.93-0.99) [b]	n.o. [a]	n.o. [a]
- Contents of information	0.81 (0.54-0.93)	0.83 (0.59-0.94) [b]	0.86 (0.63-0.95) [b]	0.81 (0.52-0.93) [b]
- Instruct	0.86 (0.64-0.95)	0.81 (0.54-0.93) [b]	0.85 (0.61-0.95) [b]	0.83 (0.57-0.94) [b]
- Provide feedback	0.78 (0.48-0.92)	0.96 (0.89-0.99) [b]	0.82 (0.54-0.94) [b]	0.87 (0.65-0.95) [b]
- Impart knowledge	0.87 (0.68-0.95)	0.91 (0.76-0.97) [b]	0.75 (0.47-0.92) [b]	0.74 (0.39-0.91) [b]
Neuromotor component				
Facilitation techniques	0.95 (0.86-0.98)	0.97 (0.90-0.99) [b]	0.97 (0.91-0.99) [b]	0.99 (0.99-1.00) [b]
- Handling	0.72 (0.37-0.89)	0.80 (0.52-0.93) [b]	0.95 (0.86-0.98) [b]	0.91 (0.76-0.97) [b]
- Pressure techniques	0.97 (0.91-0.99)	0.88 (0.69-0.96) [b]	0.93 (0.80-0.98) [b]	0.97 (0.92-0.99) [b]
- Support device	n.o. [a]	n.o. [a]	n.o. [a]	n.o. [a]
- Transition	0.86 (0.64-0.95)	0.91 (0.75-0.98) [b]	0.89 (0.69-0.96) [b]	0.87 (0.65-0.95) [b]
Sensory experience	0.85 (0.63-0.95)	0.88 (0.70-0.96) [b]	0.76 (0.42-0.91) [b]	0.78 (0.46-0.92) [b]
Passive motor experience	0.71 (0.35-0.89)	0.91 (0.77-0.97) [b]	0.70 (0.70-0.89) [b]	0.92 (0.79-0.97) [b]
Self-produced motor behavior – no interference	0.95 (0.85-0.98)	0.97 (0.93-0.99) [b]	0.82 (0.53-0.93) [b]	0.94 (0.83-0.98) [b]
Challenged to self-produced motor behavior – action continued by infant	0.86 (0.64-0.95)	0.80 (0.51-0.92) [b]	0.86 (0.63-0.95) [b]	0.86 (0.62-0.95) [b]
- Little variation	0.90 (0.74-0.96)	0.87 (0.67-0.95) [b]	0.85 (0.60-0.95) [b]	0.83 (0.57-0.94) [b]
- Large variation	0.75 (0.41-0.90)	0.82 (0.55-0.93) [b]	0.84 (0.59-0.94) [b]	0.89 (0.71-0.96) [b]
Challenged to self-produced motor behavior – with overflow into handling	0.80 (0.52-0.93)	0.88 (0.70-0.96) [b]	0.84 (0.59-0.94) [b]	0.72 (0.34-0.90) [b]
- Little variation	0.85 (0.62-0.95)	0.84 (0.59-0.94) [b]	0.86 (0.64-0.95) [b]	0.72 (0.35-0.90) [b]
- Large variation	0.72 (0.36-0.89)	0.90 (0.74-0.96) [b]	n.o. [a]	n.o. [a]
Not specified actions	0.68 (0.30-0.88)	0.84 (0.61-0.94) [b]	0.81 (0.52-0.93) [b]	0.79 (0.49-0.93) [b]
No postural support	0.93 (0.81-0.98)	0.99 (0.97-1.00)	0.97 (0.91-0.99)	0.98 (0.95-0.99)
Minimal postural support	0.78 (0.48-0.92)	0.89 (0.72-0.96)	0.77 (0.44-0.92)	0.69 (0.29-0.88)
Clear postural support	0.69 (0.31-0.88)	0.89 (0.70-0.96)	0.89 (0.71-0.96)	0.89 (0.70-0.96)
Full postural support	0.69 (0.31-0.88)	1.00 (0.99-1.00)	0.98 (0.94-0.99)	0.91 (0.74-0.97)

95% CI = 95% confidence interval, ICC = intraclass correlation coefficient
[a] Not observed
[b] $p<.05$; for definition of the various categories see Appendix

infants were challenged to explore only one movement strategy. The most frequently applied subcategory of facilitation techniques was handling. About 9% of treatment time was spent on sensory and passive motor experience. At 6 months, less postural support was provided during PT actions (Table 5.3).

The data indicated that the 75th percentile of the category facilitation techniques matched 44% of treatment time, whereas the 75th percentile of the category challenging the infant to produce motor behavior by himself/herself which is continued by the infant was at 20% of treatment time. In ten sessions, more than 44% of the treatment time was spent of facilitation techniques indicating that these sessions could be classified as NDT-like. Another 10 sessions could be classified as functional as more than 20% of treatment time was spent with the category challenging the infant to produce motor behavior by himself/herself, which is continued by the infant, leaving 22 sessions with mixed contents.

During all treatment sessions, one of the caregivers was present and observed what the physical therapist was doing. In 12 sessions caregivers acted together with the physical therapist to guide the attention of the infant. Approximately 4% of the time was devoted to family involvement and educational actions. This usually consisted of educational actions for example interfering with infant's activities. Very little treatment time was devoted to the training of caregivers on how to continue treatment strategies during daily life activities and/or in the home environment. Treatment related communication between physical therapist and caregiver was more or less equally distributed between information exchange, imparting knowledge, giving feedback, and sharing information regarding handling and the application of intervention strategies into daily routines.

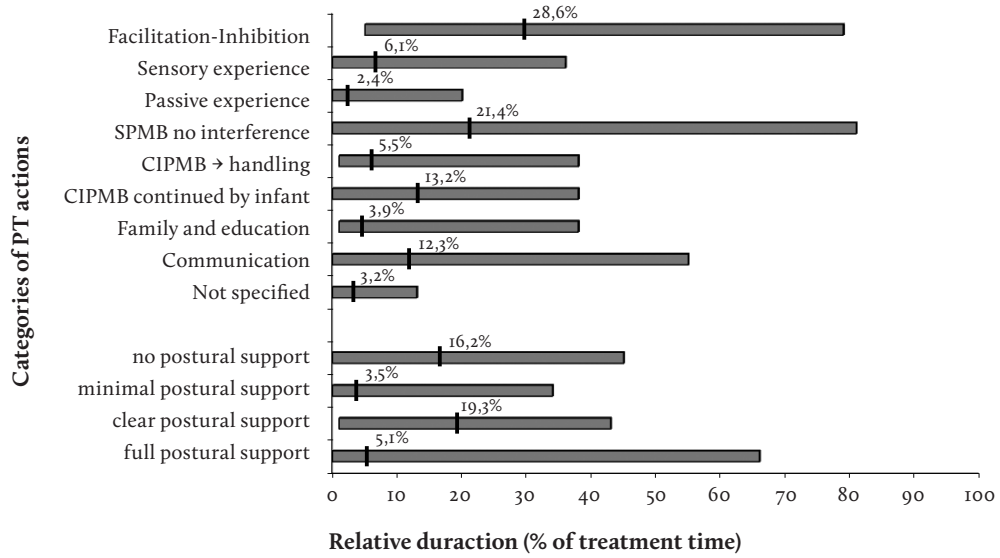
Discussion and Conclusion

The present study demonstrated that it is possible to assess PT actions during physical therapy of young infants at high risk for developmental disorders in a systematic, standardized, and reliable way. Before we address physiotherapeutic considerations, we discuss the strengths and weaknesses of our study.

One of the limitations of the study is the sample size, which results, for example, in relatively large confidence intervals for the ICC values of intra- and interrater agreement. Confidence intervals were especially large in infrequently occurring PT actions. The large confidence intervals indicate that the results cannot be generalized to any population. Therefore, it is important to realize that the results of this study should be interpreted with caution.

Another problem is that at young age CP cannot be diagnosed. This means that physical therapy at very early age is applied to infants at risk for developmental disorders, including CP, not to infants with CP. This study group was selected on the basis of definitely abnormal GMs around the age of 3 months corrected age. The presence of definitely abnormal GMs at this age is a powerful indicator of developmental disorders, including CP (Prechtl et al 1997, Prechtl 2001, Hadders-Algra 2004). Most of the infants participating in the study continued to show neurological dysfunction during the intervention period indicating they had need of ongoing physical therapy.

Figure 5.1. Relative duration (% of treatment time) of PT actions and amount of postural support (range and mean value). Results of pooling of 4 and 6 months data.



CIPMB = Challenging Infant to Produce Motor Behavior, SPMB = Self Produced Motor Behavior

The major strength of this study is that we succeeded in developing a standardized observation protocol that permitted analysis of the black box of physical therapy interventions for infants. The protocol is an instrument that may be used to assess heterogeneity in physical therapy as it offers a tool to describe operationally what therapists do in a treatment session, both across varying frames of theoretical references and across countries. The video analysis of the contents of physical therapy sessions is relatively time-consuming because it is performed on a real-time basis and usually multiple runs are needed to track down simultaneously occurring actions. Experience at our institute indicates that the technique can be learned relatively quickly and reliably. The analysis of PT actions by means of the standardized observation protocol paves the way for an objective interpretation of the contents of various pediatric physical therapy sessions.

Physiotherapeutic considerations and implications for future research

It was possible to develop an observation protocol to assess the contents of physical therapy for young infants. Eight main categories were developed that allowed the classification of single PT actions. During the developmental process, the observation protocol was checked for psychometric properties, such as completeness, mutual exclusiveness, and reliability. The results showed that the observation protocol caught virtually all PT actions during the treatment of young infants

Table 5.3. Mean relative duration of PT actions and amount of postural support at 4 and 6 months corrected age (% of time) and results of Wilcoxon signed-rank tests. Right hand column: pooling of 4 and 6 months data.

Intervention	4 mo		6 mo			Pooled data 4 and 6 mo	
	Median	range (%)	Median	range (%)	p	Median	range (%)
Family involvement and Educational component							
Educational actions	4.1	1-37	3.8	1-22	0.94	4.0	1-37
- Caregiver interferes with infant's activities	n.o.[a]		n.o.[a]				
- PT interferes with infant's activities	2.1	0-32	2.0	0-17	0.30	2.1	0-32
- PT guides the infant	0	0- 1	0.2	0-10	0.88	0.2	0-10
- PT gives caregiver training	0.1	0-13	0	0- 5	0.65	0	0-13
- PT coaches the caregiver	n.o.[a]		n.o.[a]				
Communication	10.5	0-55	15.6	0-45	0.99	12.4	0-55
- Information exchange	0.7	0-20	1.4	0-31	0.44	1.3	0-31
- Contents of information	1.0	0-16	1.4	0-14		1.5	0-15
- Instruct	0.3	0- 4	0.2	0- 4		0.3	0-4
- Provide feedback	3.5	0-17	3.1	0-11	0.85	3.3	0-17
- Impart knowledge	2.3	0-17	1.4	0-10	0.21	1.7	0-17
Neuromotor component							
Facilitation techniques	31.7	5-74	28.1	7-54	0.28	28.6	5-74
- Handling	16	4-57	16.5	3-42	0.63	16.3	3-57
- Pressure techniques	6.3	0-25	5.3	0-24	0.25	6.2	0-25
- Support device	0	0-25	0	0-15	0.27	0	0-25
- Transition	4.7	0-17	4.0	1-16	0.96	3.8	0-17
Sensory experience	8.5	1-21	6.1	0-36	0.64	6.1	0-36
Passive motor experience	4.6	0-20	2.0	0-19	0.09	2.4	0-20
Self-produced motor behavior – no interference	19.5	0-73	21.3	1-83	0.10	21.3	0-81
Challenged to self-produced motor behavior – action continued by infant	15.9	0-32	12.3	2-38	0.20	13.2	0-38
- Little variation	12.4	0-28	8.3	1-30	0.11	9.1	0-30
- Large variation	1.3	0-16	0	0-27	0.45	1.0	0-27
Challenged to self-produced motor behavior – with overflow into handling	5.5	1-25	5.6	1-37	0.03*		
- Little variation	4.9	0-23	3.7	0-37	0.04*		
- Large variation	0	0-25	0	0- 6	0.33	0	0-25
Not specified actions	1.9	0-13	3.5	0-11	0.16	3.2	0-13
No postural support	8.2	0-41	20.2	3-45	0.03*		
Minimal postural support	2.1	0-11	5.0	1-34	0.05*		
Clear postural support	13.9	2-36	22.1	1-42	0.01*		
Full postural support	8.2	0-66	4.2	0-32	0.20	5.1	0-66

a Not observed

* Wilcoxon signed-rank test: $p<.05$; for definition of the various categories see Appendix

and that it had a good inter- and intrarater reliability. The good reliability also indicated that the categories in the observation protocol were mutually exclusive. The small proportion of treatment time (3%) which could not be classified into the categories of the observation protocol mainly was spent on comforting the infant and physical therapist or caregiver and infant being out of camera view. We may thus conclude that our observation protocol is an appropriate instrument for the opening of the black box of physical therapy for infants with a high risk for developmental disorders.

The data represent the contents of various pediatric physical therapy sessions. They give an indication of how physical therapy is applied to young infants at high risk for developmental disorders in the Netherlands. The data showed that the relative duration of the applied treatment techniques covers a wide range, implying a large heterogeneity in the practical implementation of physical therapy for infants. Presumably, this does reflect reality because part of the PT actions that are performed belong to the concept of NDT and baby treatment, which by itself are well-known for its heterogeneity in application (Bly 1991). The combination with PT actions from a more functional point of view, such as the encouragement of self produced motor behavior, makes treatment application even more diverse. Presumably, the heterogeneity is brought about by the evolution of treatment techniques and theoretical assumptions over the years. First, physical therapists became aware that motor achievements occurring after the application of former treatment techniques, such as those of NDT, did not automatically carry over into activities of daily life. Nowadays, goals are defined more in relation to function, the needs of the caregiver, and new treatment approaches have been incorporated into older ones (Howle 2002). With respect to our 'Opening of the Black Box' tool, it is important to note that – despite the heterogeneity in the implementation of physical therapy for young infants – virtually all PT actions could be classified with the help of the protocol.

Across the world, treatment application in infancy varies from NDT to Vojta and from developmental interventions to stimulation programs. Thus, it would be interesting to know whether the protocol can be used also to evaluate physical therapy for infants in other countries. This study indicates that also within the application of a specific approach in one country heterogeneity is present.

Physical therapists frequently expressed surprise when they got feedback on the contents of their treatment sessions. They became aware of the discrepancy between what they believed that they had been doing and what they actually had been doing. This finding illustrates the difficulty of verbal communication. Success of verbal communication depends not only on the accuracy of the speaker or author and the intention of the hearer or reader to interpret as precisely as possible but also on the body of knowledge and the verbal frame of reference present in sender and receiver. These frames of reference may differ more often than we realize, resulting in miscommunication. Our findings suggest that the application of video feedback may serve as an instrument to prevent this type of misunderstanding. Video recording thus may be a useful tool in the training of novice pediatric physical therapists.

This study indicates that the categories challenging the infant to produce motor behavior by himself and facilitation techniques were applied most. Three types of motor behavior produced by the infant were distinguished: behavior which was initiated by the infant and behavior which was challenged by caregiver or therapist, who a) let the infant try on her/his own or b) took over control of the infant's movements by means of handling. We observed that self initiated activity of the infant usually occurred when the therapist started to communicate with the parents, leaving the infant in a situation in which his or her capacities were not challenged. Another observation was that during motor skill practice the physical therapist usually challenged the infant to explore only one movement strategy. This means that during the physical therapy sessions of this study, little variation in motor behavior was practiced. This is surprising because current concepts of infant motor dysfunction suggest that varied practice is beneficial for motor development (Hadders-Algra 2000b).

The protocol pays specific attention to family involvement and educational actions. In the latter, we distinguished between educational actions regarding the child and educational actions regarding the caregiver, that is, caregiver training and caregiver coaching. It was remarkable that during treatment, little time was spent training the caregiver on how to apply treatment strategies during daily life activities and/or in the home environment. According to the traditional treatment approach, merely based on NDT, guiding and training of caregivers are the primary technique to achieve carryover from treatment to everyday life (Bobath and Bobath 1984, Finnie 1996, Howle 2002). In the functional approaches caregivers are the key persons in the child's development and education (Law et al 1998, Rosenbaum et al 1998, Ketelaar et al 2001, Ekström et al 2005). Thus, it seems that actual practice of the therapists involved in this study differed from current notions because caregivers mainly acted as a passive observer of treatment instead of being actively involved and instructed.

Future research should aim firstly at a replication of this study in infants at various ages, with various degrees of neurological dysfunction, and in various countries. A next step will be to relate the contents of infant physical therapy sessions to developmental outcome. This knowledge might shed light on the potentially effective and ineffective elements in infant physical therapy.

In conclusion, this study demonstrated that it is possible to assess PT actions during physical therapy of young infants at high risk for CP in a systematic, standardized, and reliable way despite the heterogeneity in the implementation of infant physical therapy. The study indicated that opening the black box of infant physical therapy has shown that in the Netherlands the application is very heterogeneous and varies between more traditional (NDT-like) to more functionally oriented treatment. Our study may be seen as a first step in the development of a tool to describe therapists' actions during physical therapy interventions. Objective knowledge on the contents of physical therapy sessions is an essential step towards evidence-based practice. The next steps to be taken are replication studies on reliability assessment, studies in different populations (eg, older children with CP or developmental coordination disorder) and in different countries, and studies connecting the contents of physical therapy sessions with developmental outcome.

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CHAPTER 6

DIFFERENCES BETWEEN THE FAMILY-CENTERED PROGRAM "COPING AND CARING FOR INFANTS WITH SPECIAL NEEDS" AND INFANT TREATMENT BASED ON PRINCIPLES OF URODEVELOPMENTAL TREATMENT

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Abstract

Background. Evidence for effectiveness of pediatric physical therapy in infants at high risk for developmental motor disorders is limited. Therefore, we have developed the early intervention program COPCA: COPing with and CAring for infants with special needs – a family-centered program. COPCA is based on two components: 1) family involvement and educational parenting, 2) the neuromotor principles of the Neuronal Group Selection Theory. The COPCA coach uses principles of coaching to encourage the family's own capacities for solving problems of daily care and incorporate variation, along with trial and error in daily activities.

Objective. To evaluate whether the content of COPCA sessions differs from that of traditional infant physical (TIP) therapy sessions.

Design. Quantitative video analysis of therapy sessions with infants participating in a randomized controlled trial.

Subjects and methods. Forty-six high-risk infants were randomly assigned to receive COPCA (n=21) or TIP (n=25) between 3 and 6 months corrected age (CA). TIP in the Netherlands is generally based on NeuroDevelopmental Treatment (NDT) principles. Intervention sessions were videotaped at 4 and 6 months CA, and analyzed with a standardized observation protocol for the classification of physiotherapeutic (PT) actions. Outcome parameters were relative amounts of time spent on specific PT actions.

Results. The content of COPCA and TIP differed substantially. For instance, in TIP sessions 20 more time was spent on facilitation techniques, including handling (29% vs. 3%, p<0.001) than in COPCA sessions. During COPCA more time was spent on family coaching and education (16% vs. 4%, p<0.001) than during TIP.

Conclusion. COPCA differs broadly from TIP in terms of its NDT basis, justifying the label "novel program." Studies on the effectiveness of this family-centered program are required.

Introduction

Worldwide, therapists are trained in specialized infant treatment based on the principles of the NeuroDevelopmental treatment (NDT) approach. The original philosophy – the basic assumptions and clinical concepts of NDT in infancy – can be attributed to Karl and Bertha Bobath (Bobath 1967) and Elsbeth Köng (Köng 1966), whereas Mary Quintin was responsible for a major part of the practical implementation of the Bobath principles in infant treatment (Bly 1999). However, daily practice of the approach in different countries and healthcare settings reveals that, since then, numerous modifications and changes in the implementation of infant treatment have occurred.

Notwithstanding the occurrence of substantial changes in NDT treatment in infancy, three recent systematic reviews on the effect of early intervention in infants at high risk for developmental disorders indicated that application of NDT does not result in improved developmental outcome (Blauw-Hospers and Hadders-Algra 2005, Blauw-Hospers et al 2007, Spittle et al 2007). The reviews also indicated that best results are achieved by application of specific motor-training programs and general developmental programs.

These findings and novel insights into the biological and psychological principles governing motor development after a lesion of the brain at early age (Hadders-Algra 2000b, Hadders-Algra 2008b, Diamond 2009), along with an evolving shift in the manner in which family-centered services (FCS) are delivered, have inspired us to develop the COPCA program (Dirks and Hadders-Algra 2003). During the last few decades it has become clear that "family-centered" is a crucial aspect of interventions applied in infants and young children (Als 1992, Rosenbaum et al 1992, Dunst and Trivette 1996, McBride and Petersen 1997, Law et al 1998, King et al 2004, Rosenbaum 2004, Sanders et al 2004, Nijhuis et al 2007).

COPCA, that is, COPing with and CAring for infants with special needs, a family-centered program, is an intervention program for families of infants at high risk for developmental motor disorders after term age. The theoretical contents of COPCA differ broadly from Traditional Infant Physical Therapy (TIP) based on NDT (see Table 6.1 for an overview). Before we move on to the actual study, we should first summarize the principles of the COPCA program and TIP based on NDT.

COPCA

COPCA is rooted in the framework of the International Classification of Functioning, Disability and Health for Children and Youth (World Health Organisation 2007). It aims to promote activities and participation of the family, including the infant with special needs, while taking into account the limitations imposed by the infant's bodily impairments. COPCA consists of two components:

1. A family involvement and educational parenting component in which the key elements are: a) family autonomy in decision-making processes and family responsibility for the quality of life (Law et al 1998) and b) family-specific education, that is, the family-specific styles of parents for

educating the child to independency and personal responsibility. This means that COPCA not only respects the autonomy of the family in the process of parenting, but also values the role of the individual characteristics of both caregiver and child within the process (Scarr 2001, Sanders et al 2004).

2. A neurodevelopmental component based on the principles of the Neuronal Group Selection Theory (Edelman 1989).

The major aims of COPCA, which are listed below, are closely related to these two components.

1. The family involvement and educational component

A fundamental idea of COPCA is the transactional perspective on patterns of reciprocal contingent interactions between caregiver, siblings, and infant, along with the notion that changing patterns of behavior are a characteristic of early development (Fiese 1997, De Weerth and Van Geert 2002). This means that the family is regarded as a dynamic system of bidirectional dyads. The family itself is nested in a larger system including societal influences, socioeconomic factors, and current living conditions (Bronfenbrenner 1967). COPCA aims to encourage family responsiveness, in other words, it aims to promote the capacity of those involved in the family relationships in order to respond appropriately to each other's signals, communication, and changing states. In the case of infants with special needs, responsiveness includes the caregiver's recognition of the signals of the infant and the caregiver's responses to the actual needs of the infant, and not to preconceived ideas of what the baby might need (Mahoney 1998). To achieve these goals, the principles of coaching are used. The term "coaching" has various connotations. For instance, Als (1992) uses verbal and non-verbal coaching strategies to instruct parents in the neonatal intensive care unit on how to observe and analyze the infant's neurobehavior and to hold, handle, feed, and bathe the infant. In NDT, treatment coaching is used to teach parents special therapeutic skills such as handling. NDT provides parents with references about what the therapist is doing and furnishes parental assignments while the therapist treats the infant (Howle 2002). The therapist uses parent training to teach parents what they have to do or could do in daily care activities.

In COPCA, coaching implies the promotion of creative exploration of the competencies of the family members, including the infant with special needs, in order to stimulate self-made decisions and improve the quality of life (Hamlin et al 2009). In other words, COPCA encourages families to follow their own ideas. This is a strategy that fits the following parental expression. "Don't tell me what I can do, have to do, or must do; help me to discover it by myself." This also implies that the coach does not have an instructional role but instead supports family members on the basis of an ongoing equal partnership in order to reveal their competencies, goals, desires, and hopes. To this end, the coach uses focused bidirectional, nuanced, and detailed conversations. An important aspect of the process of coaching is the unconditional acceptance of the responsibility of the family.

COPCA coaches the family by creating a process in which the family's needs and wants are translated into solutions for how to cope with the problems related to the infant's development, while respecting the roles, culture, traditions, and routines of the family (Fiese 1997), along with

the family's own educational perspective. The coach listens, informs, and observes while the caregiver is involved in daily routines with the child, including play, thereby creating a situation in which the caregivers feel free to explore and discuss alternative strategies. Specific attention is paid to the role of siblings in care and play, along with sibling well-being. The above implies that COPCA is *not* a program in which professionals, parents, or others are engaged in "treating" the infant. COPCA is a family-centered program for professionals involved in the care of infants with special needs such as physical therapists. The aim of the program is to encourage the family's own capacities for solving the problems of daily care in naturally occurring parenting situations. Therefore, the program is delivered preferably in the home environment. Our primary target as far as professionals are concerned is pediatric physical therapists, since infants at high risk for motor disorders are generally referred to these professionals. However, other professionals, such as nurses, special educators, social workers, or occupational therapists could also become COPCA coaches. To underline the importance of coaching, we would rather call the professional applying the COPCA program a "coach" rather than a "therapist."

The problems of parenting are often complex and may vary from concerns about the infant's future locomotor development, feeding difficulties, and choice of toys, to concerns about understanding infant behavior and educational problems such as to what extent should a parent interfere with the infant's actions or whether siblings should be allowed to play with the infant. Other factors which may contribute to family distress are social issues such as time management (how to distribute time between child, siblings, partner, family, friends, work, etc.) and concerns about service delivery.

COPCA acknowledges that raising a child is a life-long challenge. Raising an infant with special needs due to an early lesion of the brain and/or neurological dysfunction adds to this challenge (Brehaut et al 2004, Raina et al 2005). Some examples of additional challenges which caregivers of infants with special needs may encounter and the way COPCA coaches deal with them are:

- The uncertain developmental of an infant with neurological dysfunction and/or an early lesion of the brain may induce parental stress (Rentinck et al 2007). Coaching addresses these uncertainties about the child's health, prognosis, and need for special services. The family members are encouraged to improve their personal coping skills.
- Stress may be associated with processes of bereavement – the mourning associated with the loss of the prospect of a typically developing child – when the chances of persisting disability are high (Kearney and Griffin 2001). Typically the process of mourning is a step-by-step process, involving cycles of feelings of disappointment, sadness, grief, anger, and guilt, which occur each time that caregivers realize that the future prospects have to be adjusted once again. COPCA focuses on discussing these processes with family members and encouraging the family to accept and express feelings of bereavement. COPCA coaches highlight the family's own strengths and capacities, and discuss the possibility of external sources of support such as relatives, neighborhood, friends, or the religious community, or the option of professional help.

- The infant's neurological dysfunction may induce atypical infant behavior which may interfere with intuitive parenting capacities (Papousek and Papousek 1984). COPCA focuses on the promotion and restoration of the intuitive parenting capacities by explaining to caregivers the mechanisms of caregiver-infant interaction and the effect of neurological dysfunction on this interaction (Sameroff and McKenzie 2003). It also stresses that parenting is a developmental process involving trial and error, in addition to life-long learning on the part of both caregiver and child (Cowan 1998). Specific attention is paid to educational actions, including play in child-preferred activities whereby the motor principles of NGST are taken into account.
- The infant's neurological dysfunction and/or an early lesion of the brain may result in atypical motor development. This may introduce the need for additional equipment such as a special chair or a supporting device for walking. COPCA coaches inform and discuss with caregivers the possible strategies and options for handling these additional needs, and it is here that the motor principles of NGST are taken into account.
- The prospect of having a child with a persisting disability introduces many practical and emotional problems into the family, problems which may vary from practical care issues for the child with special needs to dealing with service providers and organizing technical adaptations to the living environment (Raina et al 2005). COPCA focuses on coaching the family members in such a way that they are fully able to participate in the community, including the practical problems related to the presence of an infant with special needs.

In general, pediatric physical therapists are trained in addressing issues of motor development and motor dysfunction, while at the same time providing their work within the framework of family-centered philosophy (Bly 1999, Chiarello and Effgen 2006). Nevertheless, translation of the principles of family-centered services into the actual behavior of service providers in an appropriate family-centered approach may be difficult (O'Neill and Palisano 2000, Nijhuis et al 2007). This is illustrated by the findings of O'Neil and coworkers (2000) which suggest that therapists who believe in family-centered care consider the aspects of functional activities and child characteristics – reflective of a child-focused perspective – as being of paramount importance in clinical decision making. COPCA's perspective on family-centered care is to empower the family to use their own competences and problem-solving skills in the process of coping with the daily task of caring for and rearing a child with special needs. This implies a shift from family-centered care with a child focused perspective to a family-relationship-focused perspective in which the motor principles of NGST are only one of the elements taken into account

COPCA recognizes that professionals may perceive limitations in the coaching approach, since family autonomy implies limitations in terms of the extent to which educational action problems and difficulties in the community may be resolved. In those cases where the infant has complex morbidity, for instance, motor dysfunction associated with serious feeding, speech, and language problems, low vision, or in those cases where a need for additional equipment arises, the COPCA coach functions as one of the members of the interdisciplinary team involved in guiding the family.

2. *The Neurodevelopmental component: NGST*

According to NGST, typical motor development is characterized by two phases of variability: the phases of primary and secondary variability. The borders of variability are determined by genetic instructions (Edelman 1989, Hadders-Algra 2000a, 2008a). During the phase of primary variability, motor behavior is characterized by abundant variation. The motor variations represent a fundamental developmental phenomenon; they are brought about by explorative activity of the nervous system. The exploration generates a wealth of self-produced afferent information which, in turn, is used for further shaping of the nervous system. At that stage, the afferent information is not used for adaptation of motor behavior to environmental constraints. In other words, the phase of primary variability is characterized by variable but non-adaptive motor behavior.

At a certain point in time, the nervous system is able to use the afferent information produced by behavior and experience in order to select the motor behavior which fits the specific situation best; the phase of secondary or adaptive variability then starts. This selection process is based on active self-produced sensorimotor trial and error experiences. The transition from primary to secondary variability occurs at function-specific ages. For instance, in the development of sucking behavior the phase of secondary variability starts prior to term age, and in the development of foot-placing during walking it starts between 12 and 18 months (Hadders-Algra 2000a). Around the age of 18 months all basic motor functions have reached the first stages of secondary variability.

Due to the ingenious interaction between self-produced motor activities with trial and error learning and the long lasting developmental processes in the brain, it takes until the age of 18 to 20 years before the secondary neural repertoire has obtained its mature, adult configuration (Hadders-Algra 2008a).

In terms of NGST an early lesion of the brain has two major consequences (Hadders-Algra 2000b, 2008b). First, the repertoire of motor strategies is reduced. This results in less variable and more stereotyped motor behavior. In addition, infants with a prenatally, perinatally, or early postnatally acquired lesion or malformation of the brain have problems with the selection of the most appropriately adapted strategy out of the repertoire. The selection problems have a dual origin: they are related to the deficits in the processing of sensory information and to the fact that the best solution may not be available due to repertoire reduction.

The practical consequences of the above-mentioned problems are twofold. First, the limited motor repertoire may result in the absence of a specific motor strategy which would be available as the best solution in a specific situation for a typically developing infant. Due to absence of the "best" solution, the infant with an early lesion of the brain may have to choose a motor solution which differs from that of the typically developing infant. This implies that the different motor behavior of an infant with an early lesion of the brain should not a priori be regarded as deviant, that is, as something which deserves to be "treated away," as it may be the infant's best and most adaptive solution for the situation (Latash and Anson 1996). Second, due to the deficits in the processing of sensory information which hamper the process of selection of the best strategy, infants with an early lesion of the brain need ten- to a hundredfold more active motor experience

than typically developing children (Valvano and Newell 1998).

Translating NGST principles into daily practice of the COPCA program means that the COPCA coach will inform and coach family members while taking into account the family's educational perspective. Family members receive suggestions for incorporating variation and trial and error in daily activities, since this might enhance the infant's motor repertoire and promote the ability to select the best strategy for different conditions. This also implies that COPCA is characterized by being "hands-off." Some examples of variation in activities are those variations in which the infant is challenged to produce self-initiated motor behavior in natural daily situations such as playing, bathing, or sitting. In addition, suggestions are provided about various ways the infant may be encouraged to explore the limits of its growing capacities. Interestingly, typical play with siblings usually is an excellent means of promoting sensorimotor challenge, variation, and exploration. At the same time, family members are informed about the basic principles of atypical development. The guidelines are: a) the notion that the appearance of motor behavior of the child with special needs may differ from that of a typically developing child, but that this does not matter as it is functional performance that counts; and b) the idea that development implies trial and error during self-produced activity, in other words, that "error" production does not mean failure but the presence of an active learning process, where learning may be enhanced by verbal encouragement or non-verbal expressions of affection (e.g., hugging, touching, or tickling) by the caregiver. The notions of NGST are not taught but discussed by means of bi-directional, equivalent, and deliberate conversation, in line with the family involvement and educational component of COPCA.

TIP based on NDT

TIP based on NDT is a multidisciplinary approach which primarily focuses on limitations imposed by bodily impairments and functional activities of the infant with special needs. The assumptions of the approach can be summarized with the help of these two major components:

1. Neurodevelopmental principles which consist of a mix of neuromaturational assumptions, sensorimotor problem-solving strategies resembling those based on NGST, and the principles of dynamic systems. The therapist plays a key role in the application of these principles due to her/his instructional qualities of specific handling techniques and experiences. The therapist teaches the infant to engage in developmental activities by providing sensorimotor experiences accompanied by manual guidance, toys, a bolster, or a ball (Bly 1999, Howle 2002).
2. The family. The role of the family in the planning of the treatment program has been eloquently expressed by Bly (1999): "The family members are obviously the most important people on the baby's team" and "The more involved the family becomes, the more consistent therapeutic management becomes for the baby" (p. 8).

   The major aims of TIP based on NDT, which are listed below, are closely related to these two components.

1. *The neurodevelopmental component*

In NDT the understanding of how typical developmental changes across a lifespan provides a critical framework for the problem-solving approach and the planning of treatment. Development is understood as an interplay between endogenous maturational processes and the infant's own environment. NDT uses problem solving to identify missing or atypical elements of functional movements and posture in infants with a prenatally, perinatally, or early postnatally acquired lesion or malformation of the brain (Howle 2002).

The process of the guidance of an infant starts with the examination of functional limitations. Next, on the basis of proposed relationships between limitations, intervention strategies are systematically planned and applied. NDT emphasizes the individual as a whole with his/her own competencies and limitations. The current enablement model of NDT addresses these competencies and limitations – in line with the ICF-CY – in body, motor, individual, and social dimensions. In infant treatment NDT emphasizes impairment as the starting point of therapeutic guidance and improvement of impairment as a means to facilitate activity and participation (Bly 1999). According to Mayston (2001) the basic philosophy underlying the sensorimotor deficits of the infant with a brain lesion are pathophysiological insights into the mechanisms underlying the problems in the coordination of posture and movement combined with atypical qualities of muscle tone. The problems contribute directly to functional limitations. The impairments of function can be classified into primary impairments, that is, those impairments that result directly from the neuropathology along with secondary impairments. Two forms of primary impairments can be distinguished: positive impairments such as spasticity and deficits in postural control, and negative impairments such as muscle weakness and changes in muscle length. Those impairments that are associated with atypical interactions within and between neural and body systems and other environmental influences, such as contractures and atrophy, are classified as secondary impairments (Howle 2002). For the implementation of NDT in daily practice this means that a broad repertoire of tone-influencing facilitation techniques such as handling are used to reduce atypical functional activities and to prepare the infant for optimally independent function.

Bly (1999) emphasized the importance of tactile, proprioceptive, kinesthetic, and vestibular sensory input and the integration with the motor system in infant treatment based on NDT. The sensory input enables the infant to perform movements and postural adjustments, and to learn and use motor skills. Therefore, assessment of the infant must include attention to the sensory system and treatment must include techniques that integrate sensory input with motor control. The forms of NDT able to achieve this most precisely are the facilitation techniques of handling, intermittent pressure, and tactile and vestibular stimulation. The hands of the therapist are used to control sensory input, motor output, and muscle tone in such a way that the infant with neurological dysfunction gains an experience of typical movements and movement sequences with the aim of enhancing the infant's development of learning skills (Bly 1999).

At the same time the approach recognizes that typical development of postural control and movements relies on the concept of dynamic systems (Thelen et al 1993). According to dynamic sys-

tems, motor development is brought about by self-organization; it is a result of interaction between multiple factors such as body weight, muscle strength, joint configuration, the infant's mood, and the environment, including the characteristics of the task. These variables change during the development of the infant, and NDT uses these variables to influence outcomes, for instance, by adapting the intervention to the age of the infant and the infant's characteristics in a direct teaching relationship with the infant (Mayston 1992, Howle 2002). This implies that the therapist should treat the infant and, during hands-on treatment, should select handling strategies that facilitate and prepare the infant for age-specific function. The caregivers are instructed on how to continue and integrate these treatment strategies which often continue to involve hands-on techniques throughout daily life.

2. *The family involvement component*

Therapists, other professionals, and the caregiver act like a team in the decision-making process in order to establish the infant's care plan. The role and responsibility of each discipline is described in the treatment plan. This means that the therapy disciplines need to develop a treatment plan in which the therapist and caregiver cooperate in the formulation of meaningful and relevant functional goals. The therapist presents options and discusses these options with the parent in order to reach a consensus, while anticipating the goals that the infant might achieve. The responsibility of the therapist is to discover the best way for the infant to achieve his/her best potential. This implies that the therapist should teach the child and train the caregivers and other family members in the most useful ways of managing the infant (Bly 1999, Mayston 2001, Howle 2002).

The treatment plan incorporates caregiver education which is intended to facilitate the caregiver-infant relationship, teach caregiver-specific skills, and enable the caregiver to handle/assist with their child's difficulties. This means that it is the responsibility of the caregiver to carry out specific treatment strategies and to provide opportunities for practice. It is the therapist's role to guide the caregiver in how to modify the normal care-giving activities so that each daily task can be used to reinforce the improvement of motor patterns which the infant has learned during the therapy session. A greater involvement by the caregiver will express itself in terms of a more consistent therapeutic management of the infant. In order to achieve an optimal result, the therapist informs, instructs, and may give caregivers as many treatment ideas as the caregiver can manage (Finnie 1996, Bly 1999). In addition, in TIP based on NDT, communication is regarded as an essential means for service delivery. Communication is characterized by an open information exchange between a source of information (the professional) and a receiver (the caregiver). In this way information is exchanged about, for instance, specific details of the diagnosis, the child's prognosis, as well as on the presence or absence of physiological reactions, changes in muscle tone, "why" and "how" to use handling techniques, along with communication skills (Finnie 1996).

The aim of the present study is twofold: 1) to examine whether it is possible to implement the concepts of COPCA in daily physical therapy practice by specially trained pediatric physical therapists and 2) to evaluate whether the content of COPCA sessions differs from TIP sessions.

Table 6.1. Overview of differences between COPCA and TIP based on NDT.

	COPCA Family-relationship focused	**TIP based on NDT** Child focused
Theoretical frameworks - Family involvement - Family education - Child development - Education of the infant	- Family autonomy (Law et al 1998) - Transactional model (Sameroff and Mackenzie 2003) & ecological model (Bronfenbrenner 1967) - NGST (Edelman 1989) - Individuality in parenting (Scarr 2001, Sanders et al 2004)	- Family centered care guided by professional expertise (Howle 2002) - Professional-client didactic relationship (Howle 2002, Mayston 1992) - Mix of neuromaturation, dynamic systems and NGST (Howle 2002) - General parenting principles (Finnie 1996)
Primary focus of guidance	- Decision making process of the family	- Optimizing child development
Role of family	- Key factor in process of coaching	- Member of the team
Role of therapist	- Coach	- Key person in guidance - Teacher
Education of infant	- Coaching of caregiver - Educational perspective	- Therapist teaches infant and trains caregiver
Key words of motor learning	- Variation - Self-exploration - Challenge, testing the limits - Trial and error - Acceptance of atypical strategies	- Exposure to sensory-motor experience - Facilitation of typical motor behavior, avoidance of atypical behavior - Hands on → hands off techniques
Communication	- Bidirectional, equal partnership and open dialogue	- Open information exchange between a source of information (the professional) and a receiver (the caregiver).

Method

Subjects

Forty-six infants, who had been admitted to the Neonatal Intensive Care Unit (NICU) of the University Medical Center Groningen between March 2003 and May 2005 and who presented at the age of ten weeks corrected age with definitely abnormal general movements (GMs), were included in the study. GM assessment was carried out by MH-A and neonatologist AFB, who agreed on the infant's GM quality. The presence of definitely abnormal GMs indicates a high risk for developmental disorder, including CP (Hadders-Algra 2004). Infants with severe congenital anomalies and infants whose caregivers had insufficient understanding of the Dutch language were excluded from the study. All caregivers of the infants signed an informed consent and the research project was approved by the Ethics Committee of the University Medical Center Groningen. The trial was registered under ISRCTN85728836.

Block randomization (full-term infants blocks of n=2; preterm infants blocks of n=12) allocated 21 infants to the experimental COPCA group and 25 infants to the control group. The infants who were randomized into the control group received traditional infant physiotherapy (TIP) on indication of the pediatrician. As a result, 22 infants were referred to TIP and three infants did not receive physical therapy. Each group contained two full-term infants; the other infants had been born preterm at gestational ages which varied from 25 to 36 weeks. Details of perinatal and social characteristics of both groups are provided in Table 6.2.

The randomized intervention was provided at between 3 and 6 months corrected age (CA). The COPCA sessions were performed twice a week for one hour in the home environment by one of four COPCA coaches. The COPCA coaches were certified and registered pediatric physical therapists who had received specific education in the COPCA program. The educational curriculum of a COPCA course starts off with two days of training in the basic COPCA principles. These two days include discussions of videos of recent intervention sessions carried out by participants. Discussion of the videos allows for clarification of key characteristics of COPCA and the differences between COPCA and TIP. After the introductory two days, training is continued with four one-day sessions with an interval of six weeks. During the interval, coaches-in-training videotape their own intervention activities. Again, the videos are the starting point for discussion of how the principles of COPCA may be applied in daily practice. The COPCA curriculum also involves obligatory reading. During the study, COPCA coaches continued to receive supervision from the first author. In earlier phases of their careers, three of the four COPCA coaches had completed the NDT basic and infant course based on Neurodevelopmental Treatment (NDT) principles. Seventeen pediatric physical therapists were involved in the TIP treatment sessions. Sixteen of them had completed the NDT basic and infant treatment course based on NDT. The frequency of TIP sessions varied from two to 28 times (median 9), and their duration from 12 to 50 minutes (median 29). The great majority of TIP sessions were also carried out in the home environment (n=20; 91%) Therapists for both groups had at least five years experience in treating infants and children with special needs. None of the infants received additional forms of therapeutic guidance.

Table 6.2. Perinatal and social characteristics of both study groups.

		COPCA (n=21)	TIP (n=25)
Gender			
▪ Male	n (%)	9 (43)	11 (44)
▪ Female	n (%)	12 (57)	14 (56)
Gestational age at birth (wk)	median (range)	29 (27-40)	30 (25-39)
Birth weight (g)	median (range)	1210 (585-4750)	1143 (635-3460)
Maternal age	mean ± SD	30.5 ± 6.2	31.8 ± 4.3
Firstborn child	n (%)	12 (57)	13 (52)
Twin pairs	n (%)	9 (43)	7 (28)
Abnormal cerebral ultrasound			
▪ IVH gr. 3-4	n (%)	1 (5)	1 (4)
▪ PVL gr. 3-4	n (%)	2 (10)	1 (4)
Cerebral Palsy at 18 months	n (%)	5 (24)	5 (20)
Maternal education *			
▪ Low	n (%)	3 (14)	3 (12)
▪ Middle	n (%)	16 (76)	11 (44)
▪ High	n (%)	2 (10)	11 (44)

* high vs middle + low education: Chi square: p<0.05

SD = standard deviation; wk = weeks; g = gram; gr = grade; IVH, grading according to Volpe (2000) = intraventricular haemorrage; PVL, grading according to de Vries et al. (1992) = Periventricular leukomalacia; Levels of education: low = primary education/ junior vocational training, middle = secondary education / senior vocational training, high = university education / vocational colleges.

Video recording and video analysis

At 4 and 6 months CA, a video recording of a physical therapy session was made. To minimize intrusion, the camera was positioned as far away from the therapist and infant as possible. Albrecht and coworkers (2005) demonstrated that video recording scarcely affects the behavior of the people being filmed. In the COPCA group, one recording was missing at four months and three at six months due to logistical difficulties. In the TIP group, one recording was missing at four months (logistics) and one at six months (treatment had already stopped). This resulted in 41 video recordings at four

months (COPCA n=20, TIP n=21) and 39 recordings at six months (COPCA n=18, TIP n=21).
Two persons, CBH and LH, analyzed the contents of the sessions with a standardized observation protocol for the classification of PT actions (Blauw-Hospers et al 2010). Both assessors were blind to group allocation. The analysis was performed with the Noldus software program "The Observer" (Version 5.0; Noldus, Wageningen, the Netherlands), a program specifically designed for behavioral observation. The program allows for the quantification of the duration, frequency, and serial order of defined actions. The therapeutic actions defined in the protocol reflect the specific components of COPCA and TIP (see Appendix 1, p. 188). The observation protocol classifies PT actions into eight main categories: A) Family involvement and educational actions; B) Communication, C) Handling techniques; D) Sensory experience; E) Passive motor experience; F) Self-produced motor behavior, no interference; G) Challenge to self-produce motor behavior where infant is allowed to continue activity; and H) Challenge to self-produce motor behavior that flows over into therapeutic handling. In addition, the position of the infant and the amount of postural support the infant received during the PT actions was scored. The majority of outcome parameters resulted in relative amounts of time spent on PT actions. Items from Sections 1 to 3 of Category A (Family involvement and educational actions, A1 to A3) were categorical data. Categories A and B describe aspects of family involvement and educational interaction in the intervention programs. In Categories C to H, the position and postural support parameters deal with the neuromotor components of treatment.

Previously we demonstrated that inter- and intra-assessor agreement on assessment with the protocol were satisfactory. The ICCs on the relative duration of actions ranged from 0.76 to 1.00 for inter-assessor agreement and 0.69 to 0.99 for intra-assessor agreement (Blauw-Hospers et al 2010).

Data analysis

Statistical analyses were performed using the computer package SPSS (version 14.0). Non parametric statistics were used since none of the variables were normally distributed. The effect of age was assessed with the Wilcoxon signed rank test. Differences between COPCA and TIP were evaluated with the Mann-Whitney U test, the Chi-square test or Fisher Exact test. Differences with a p-value <0.05 were considered statistically significant.

Results

Family involvement and educational components of intervention

The caregivers were involved in both types of intervention, albeit in different ways. During TIP sessions, caregivers were engaged especially in handling, and during COPCA, in playing (Table 6.3). The difference in approach between COPCA and TIP was also reflected in the way the child was dressed: during TIP, infants were more often undressed than during COPCA, a difference which reached statistical significance at four months (Table 6.3). In both types of intervention, mothers were usually present during treatment sessions.

Table 6.3. Family involvement during COPCA and TIP sessions.

Intervention	4 months COPCA n= 20	4 months TIP n= 21	6 months COPCA n= 18	6 months TIP n= 21
Family involvement				
A.1 Family members involved in intervention session				
- Mother present only	13	13	15	11
- Father present only	1	1	2	0
- Both caregivers present, no other family members	4	3	1	3
- In addition to parent(s) other family members present [1]	2	4	0	7
A.2 Role of parent, caregiver[2]				
- Physical therapist performs treatment by means of handling	1	15	0	15
- Physical therapist performs treatment (handling) and parent guides attention of infant	0	5	0	5
- Parent performs treatment by means of handling. Physical therapist instructs how to handle	0	1	0	1
- Parent and physical therapist act together (hands off); parent is playing with the infant; physical therapist observes parent –infant relationship; parent gives ample opportunities for exploration	13	0	14	0
- Parent is playing with infant (hands-off), ample opportunities for exploration	6	0	4	0
A.3 Infant dressing[3]				
- Dressed	15	5	9	6
- Partially dressed	4	5	6	5
- Undressed (wearing underwear only)	1	11	3	10

1 In the current study other persons present were always grandparents.
2. Role of parent, caregiver: small (first three categories) vs large (last two categories). Chi Square p<0.001.
3. Infant dressing: Chi Square for trend p<0.001.

Table 6.4. Contents of TIP and COPCA intervention at 4 and 6 months corrected age: proportion of time spent on various actions.

Intervention	TIP						COPCA					
	4 mo		6 mo		Pooled 4 and 6 mo		4 mo		6 mo		Pooled 4 and 6 mo	
	med	**range (%)**	**med**	**range (%)**	**med**	**range (%)**	**med**	**range (%)**	**med**	**range (%)**	**med**	**range (%)**
A. Family involvement and Educational component												
A.4 Educational actions	4.1	1-37	3.8	1-22	4.0	1-37	17.0	0-44	12.7	0-36	16.0**	0-44
- Caregiver interferes with infant's activities	0.0	0-1	0.0	0-2	0.0	0-2	**0.3**	0-3	**0.9**	0-5	0.6**	0-5
- PT interferes with infant's activities	2.1	0-32	2.0	0-17	2.1	0-32	0.4	0-8	0.2	0-7	0.2**	0-8
- PT guides the infant	0.0	0-10	0.2	0-10	0.2	0-10	0.0	0-7	0.0	0-0	0.0**	0-7
- PT gives caregiver training	0.1	0-13	0.0	0-5	0.0	0-13	0.0	0-3	0.0	0-1	0.0	0-3
- PT coaches the caregiver	0.0	0-1	0.0	0-1	0.0	0-1	12.4	0-43	11.3	0-35	11.9**	0-43
B. Communication	10.5	0-55	15.6	0-45	12.4	0-55	17.6	0-50	14.7	2-52	15.7	0-52
- Information exchange	1.6	0-16	1.3	0-14	1.4	0-16	1.8	0-9	0.9	0-9	1.3	0-9
- Contents of information												
- Developmental education	0.7	0-20	1.4	0-31	1.3	0-31	1.0	0-20	2.1	0-31	1.3	0-31
- Handling	0.4	0-10	0.3	0-14	0.4	0-14	0.0	0-1	0.0	0-0	0.0**	0-0
- Variation	0.0	0-1	0.3	0-14	0.0	0-1	1.6	0-9	0.5	0-8	1.3**	0-90
- ADL handling	0.0	0-10	0.0	0-5	0.0	0-10	0.0	0-0	0.0	0-0	0.0**	0-0
- ADL variation	0.0	0-0	0.0	0-0	0.0	0-0	0.0	0-2	0.0	0-7	0.0**	0-7
- Instruct												
- Assign	0.2	0-3	0.2	0-3	0.2	0-3	0.9	0-12	1.0	0-6	1.0**	0-12
- Give hints	0.0	0-4	0.0	0-1	0.0	0-4	**2.5**	0-11	**2.1**	0-5	2.3**	0-11
- Provide feedback	3.5	0-17	3.1	0-11	3.3	0-17	3.7	0-14	5.1	0-13	4.5	0-14
- Impart knowledge	2.3	0-17	1.4	0-10	1.7	0-17	**2.8**	0-14	**1.7**	0-9	1.7	0-14
Neuromotor component												
C. Facilitation techniques	31.7	5-74	28.1	7-54	28.6	5-74	2.6	0-43	3.3	0-11	2.9**	0-43
- Handling	16.0	4-57	16.5	3-42	16.3	3-57	0.4	0-32	0.3	0-4	0.4**	0-32
- Pressure techniques	6.3	0-25	5.3	0-24	6.2	0-25	0.0	0-5	0.0	0-8	0.0**	0-8
- Support device	0.0	0-25	0.0	0-15	0.0	0-25	0.0	0-5	1.8	0-5	0.0**	0-5
- Transition	3.6	0-17	4.0	1-16	3.8	0-17	1.8	0-5	0.0	0-1	1.8**	0-5
D. Sensory experience	8.5	1-21	6.1	0-36	6.1	0-36	1.9	0-11	1.7	0-7	1.8**	0-11
E. Passive motor experience	4.6	0-20	2.0	0-19	2.4	0-20	0.0	0-4	0.0	0-1	0.0**	0-44
F. Self-produced motor behavior – no interference	21.3	0-43	21.3	1-83	21.3	0-81	41.0	2-71	52.5	7-69	44.3**	2-71
G. Challenge to self-produced motor behavior – action continued by infant	15.9	0-32	12.3	2-38	13.2	0-38	35.2	3-81	30.8	17-70	31.0**	3-81
- Little variation	12.4	0-28	8.3	1-30	9.1	0-30	10.7	0-34	2.4	0-41	6.5	0-41
- Large variation	1.3	0-16	0.0	0-27	1.0	0-27	26.5	2-59	25.5	7-37	25.8**	2-59
H. Challenge to self-produced motor behavior – with overflow into handling	**5.5**	1-25	**5.6**	1-37	5.6	0-37	0.0	0-6	0.0	0-5	0.0**	0-6
- Little variation	**4.9**	0-23	**3.7**	0-37	4.9	0-37	0.0	0-6	0.0	0-1	0.0**	0-6
- Large variation	0.0	0-25	0.0	0-6	0.0	0-25	0.0	0-1	0.0	0-4	0.0	0-4
I. Unspecified actions	1.9	0-13	3.5	0-11	3.2	0-13	2.9	1-9	3.1	1-9	3.1	1-9

Bold indicates a significant age-effect, Mann Whitney, $p < 0.05$. Differences between TIP and COPCA ** $p < 0.01$
ADL = activities of daily life, Med = Median, Mo = months (corrected age), PT = physical therapist.

The time spent on various educational actions varied widely (Table 6.4 and Figure 6.1). Age had only a minimal effect on family involvement and educational actions; we therefore pooled the four- and six-month data for the evaluation of the differences between COPCA and TIP (Table 6.4). During COPCA sessions more time was spent on educational actions (median value 16%) than during TIP sessions (median value 4%, p<0.001). Not only did the duration of educational actions differ between the two treatments, but the contents also did. During TIP sessions, educational actions more often consisted of actions during which the therapist interfered with the infant's activities, that is, corrected, interrupted, or assisted infant activities. During COPCA sessions, most of the time was spent on coaching the caregivers, whereas this form of guidance did not occur in the TIP group (p< 0.001).

In both groups an equal amount of time was spent on communication. However, differences were present in the contents of the communication, especially in the type of information provided by the therapist (Table 6.4 and Figure 6.1). During TIP sessions, the focus was on explaining facilitation and the use of handling techniques in terms of hand placing, tone influencing, asymmetry, symmetry, and typical movement patterns. During COPCA sessions, the emphasis was on explaining the importance of variation and the role of minimal support, exploration, and trial and error. COPCA coaches more often used the instructional strategy of "giving opportunities" than did TIP therapists.

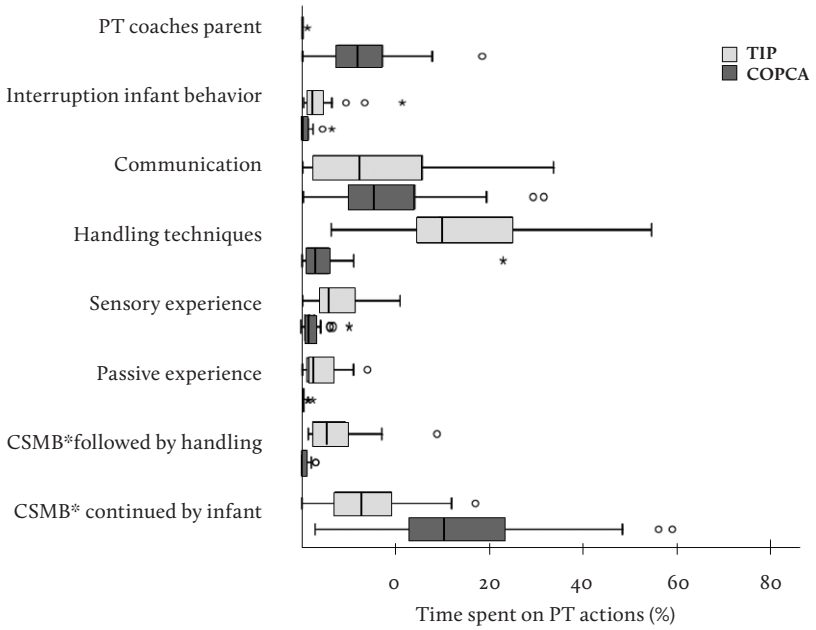

Figure 6.1: Data on the relative duration of time spent on eight categories of PT actions during treatment sessions. The horizontal lines represent ranges; the boxes, the interquartile ranges; and the vertical bold bars, median values.
CSMB = Challenged Spontaneous Motor Behavior

Neuromotor components of intervention

The application of physical therapy at four and six months was characterized by heterogeneity (Table 6.4). The relative duration of the majority of PT actions in both groups at four months did not differ significantly from that at six months (Table 6.4). The only exception to this rule was a slight increase for "Challenge to self-produce motor behavior with overflow into therapeutic handling" in the TIP group from four to six months. In light of the minimal effect of age, we considered it justified to pool the data of four and six months in our evaluation of the differences between COPCA and TIP (Table 6.4 and Figure 6.1).

During TIP sessions most of the time was spent on facilitation (median value 28%), especially on handling. Handling techniques were almost absent in the COPCA group, meaning that the difference in time spent on handling techniques between TIP and COPCA sessions was highly significant (p<0.001). During TIP sessions a substantial amount of time was also spent on granting the infant time to produce motor behavior by itself (either entirely spontaneously: 21%; challenged: 13%). During COPCA sessions even more time was spent on granting the infant time to produce motor behavior by itself, either entirely spontaneously (44%; difference with TIP: p<0.001) or challenged (31%; difference with TIP: p<0.001). We noted also a qualitative difference between the two groups during the periods of entirely spontaneous activity. In the TIP group, this meant that the infant was left unattended while therapist and caregivers were engaged in communication; in the COPCA group, this meant that caregivers and PT observed and discussed the infant's activities. When the infants during COPCA sessions were challenged by toys or the face of the physical therapist or caregiver to explore their own movement possibilities, considerably more variation was used than during TIP sessions (median values COPCA 26% vs. TIP 1%, p<0.001). During COPCA sessions little time was spent on sensory stimulation, passive experiences, and challenging of motor behavior flowing over into handling techniques; these actions occurred significantly less often than during TIP sessions (Table 6.4 and Figure 6.1).

The position of the infant during treatment varied with treatment type and age. With increasing age, infants were less frequently treated in supine position and more often in prone and sitting positions (Table 6.5). During TIP sessions, infants were handled significantly more often in supine position with the pelvis slightly lifted and in prone than during COPCA sessions; this was true for both ages. During COPCA sessions, infants were more often placed in sitting position, particularly at four months. It is well known that young infants need postural support during sitting. It was therefore interesting to note that during COPCA more time was especially spent on sitting with minimal support (Table 6.5).

Table 6.5. Infant position and degree of postural support in sitting during TIP and COPCA sessions: proportion of time spend in various positions.

Intervention	**TIP**					**COPCA**				
	4 mo		**6 mo**			**4 mo**		**6 mo**		
	Median	Range (%)	Median	Range (%)	Effect age; P-value	Median	Range (%)	Median	Range (%)	Effect age; P-value
Position										
Supine pelvis not lifted	38.9	2-66	27.9	5-60	0.06	45.6	7-92	34.3	0-55	0.68
Supine with pelvis slightly lifted	16.5	0-47	9.5	0-32	0.21	0.0 **	0-27	0.0 **	0- 7	0.06
Prone	**15.6**	7-35	**29.4**	12-43	0.002	**5.6** **	0-29	**14.2** *	0-38	0.008
Side	5.6	0-43	4.0	0-15	0.20	3.8	0-27	5.5	0-33	0.95
Sitting	**14.5**	3-83	**25.3**	9-50	0.002	36.7 **	0-72	44.9 **	5-85	0.28
- Sitting with no support	0.0	0-0	0.0	0-2	0.047	**0.0**	0- 5	**0.4** *	0-15	0.002
- Sitting with minimal support	**0.3**	0-9	**3.5**	0-32	0.0005	13.6 **	0-42	19.8 **	1-48	0.14
- Sitting with clear support	5.9	0-32	11.6	0-41	0.78	17.6 *	0-38	10.2	1-26	0.23
- Sitting with full support	7.0	0-42	4.0	0-32	0.59	1.4	0-25	1.4	0-34	0.52

Bold indicates a significant age-effect, Mann-Whitney p< 0.05. Differences between COPCA and TIP: * p< 0.05, ** p< 0.01
Mo = months (corrected age)

Discussion

The present study demonstrated that the contents of COPCA and TIP sessions differed widely, suggesting that it is possible to implement COPCA in daily practice by pediatric physical therapists who have received special training in the COPCA program. The differences found correspond to the theoretical frameworks of the two approaches. They included differences in the approach to families, the role of the caregiver and pediatric physical therapist, the application of educational actions towards the infant, and in activities to stimulate the infant's sensorimotor development.

Physiotherapeutical considerations

Our data clearly demonstrated that, notwithstanding the fact that both TIP and COPCA are defined as family-centered services (Rosenbaum 2004), important differences between the two approaches exist in application of the key element "family involvement." The videos of the COPCA sessions illustrated that family members, including the infant with special needs, were the key persons in guidance which consisted of coaching. Caregivers determined the focus of the guidance. The COPCA coaches created a situation in which caregivers felt free to explore and discuss alternative strategies. Caregivers took care of or played with the infant (in a hands-off manner) to provide the infant with ample opportunities for self-exploration. In contrast, TIP therapists usually treated the infant by using handling techniques (hands-on/hands-off) and provided parents with references about what the therapist was doing or what a parent could do in daily practice. It was also observed that TIP therapists were often in charge of decision-making. This was reflected, for instance, in the way they taught infants the "best" way to accomplish self-initiated movement goals. The observed characteristics of TIP correspond with the descriptions of baby treatment based on NDT principles (Bly 1999).

The differences between COPCA and TIP reflect a profound difference in the role of the family in the decision-making processes involved in the care of a child with special needs. During the past few decades, understanding of how to manage the care of children with special needs has changed substantially. This has resulted in the advocacy of a family-centered instead of a child-focused approach (Rosenbaum 2004). Our data on the daily practice of TIP therapists indicate that therapeutic guidance in general is still characterized by child-focused care with a unilateral dominance of the professional determining the goals of and actions in treatment. Thus the TIP approach corresponds to the direct didactic teaching principles of NDT, put into words by Mayston (1992) in the following way (p. 6): "Our responsibility as therapist is to discover the best way for our clients to achieve their best potential." The discrepancy between current theory (family focus) and daily practice is in line with the findings of O'Neil and Palisano (2000) which indicate that therapists who acknowledged the importance of family-centered care focused their professional attention on the child's impairment. We suggest that the critical determinant for family-centered services, as suggested by COPCA, is family autonomy. This implies that professionals should step back and restrict their role to coaching. The COPCA coach helps caregivers to discover

and develop their own problem-solving strategies for coping with and caring for the infant with special needs. The important distinctions between COPCA and TIP are that 1) in COPCA the role of the family is not restricted to parental involvement in the decision-making in terms of functional treatment goals, and 2) COPCA refrains from teaching caregiver intervention techniques (Bromwich 1976, Affleck 1982, Mahoney 1998).

The differences observed in activities to stimulate the infants' sensorimotor development between the two groups correspond to the theoretical frameworks of the two approaches. The videos demonstrated that the COPCA approach creates a rich and varied world of opportunities allowing the infant to explore variable motor behavior (hands-off). A case in point is the challenge of postural behavior by the provision of as little support as possible. By creating a challenging environment, the infant may, through active trial and error, experience which motor behavior is the best he or she can manage. This corresponds to the ideas of NGST. Preliminary results of a study on the effect of COPCA and TIP on postural development in early infancy suggest that COPCA may enhance the selection of a functional postural strategy (De Graaf-Peters et al 2007). The TIP approach also aims at facilitating motor behavior produced by the infant her/himself; however, our study indicated that TIP therapists frequently incorporated techniques of handling and carefully graded stimulation (hands-on). Therapists in the TIP group also applied the basic concept of NDT to prepare the infant's motor capacities for a specific function (Mayston 1992). This was, for instance, reflected in the use of treatment in supine position with the pelvis slightly lifted during TIP sessions, whereas this type of positioning was virtually absent during COPCA sessions. This specific handling technique aims to facilitate functional activities such as rolling or reaching and grasping in the midline, while the therapist controls muscle tone. It should be noted, however, that the question of how increased muscle tone is related to activity limitations and postural control is a matter of debate (Damiano 2006, Brogren-Carlberg and Bower 2008). Analogous to the findings in the family-care domain, our findings in the neuromotor domain match those of O'Neil and Palissano (2000): Despite the presence of a shift to more functional therapy, therapists generally pay more attention to movement quality than to functionality so as to achieve a better long-term outcome. This suggests that the "living concept" approach of NDT – as recognized by senior NDT tutors – may benefit from a paradigm shift from "quality" (how) to "functionality" (what) (Mayston 1992).

Methodological considerations

The major strength of the study is the standardized, video-based analysis of the contents of the two forms of infant physical therapy that allows for an objective quantification of differences between the two approaches. Other strengths of the study lie in the identical age of the infants at inclusion in the study, the identical ages at video-assessment, and identical limitations at study entry, that is, the presence of definitely abnormal GMs. The presence of definitely abnormal GMs at three months post-term is associated with a high risk of developmental motor disorders such as cerebral palsy (Prechtl 2001, Hadders-Algra 2004).

It may be considered a weakness that the therapists who applied COPCA had received a specific COPCA training shortly before the study started, whereas training of the therapists who applied TIP had taken place at longer and more variable periods prior to study onset. Nevertheless, the great majority of TIP therapists received certified NDT training including an infant treatment course. It is well known that therapists practicing NDT, develop a personal, eclectic approach as their experience increases (Mayston 2008). This was reflected in the variation in percentage of time spent on specific actions during the treatment sessions – a variation which partly blurred the contrast between TIP and COPCA. Yet, by using a control group of therapists applying regular TIP, we were able to demonstrate that the contents of COPCA sessions differed from currently practiced infant physiotherapy in the Netherlands.

The difference in treatment frequency may be regarded as another limitation, as frequency of treatment has been associated with outcome (Tsorlakis et al 2004).
At the time of the study, the current design had the highest level of feasibility. In our analysis of the effect of intervention, we took frequency of treatment into account (Blauw-Hospers et al, submitted for publication).

Infant physical therapy differs across countries and varies with the cultural context of families. This means that the findings of the present Dutch study cannot immediately be generalized to other countries.

The randomized design of the study was associated with a high degree of similarity for both groups of infants (Table 6.2). Despite randomization, the level of maternal education differed between the two groups. This difference may be considered a weakness of the study, since contents of a treatment session may be affected by the level of maternal education.

Concluding remarks

Bertha and Karl Bobath were well aware of the possibility of future progress in ideas on family care and motor development. They encouraged therapists to incorporate new ideas into daily application of therapy. In a way, one could say that the authors of the COPCA program followed the advice of the Bobaths. They developed a new program, COPCA. The current study indicates that COPCA and TIP based on NDT differ widely, justifying the label "novel program."

Our study underscores the notion that the application of standardized, computer-based video analysis of treatment sessions is an invaluable tool in the understanding of the daily practice of pediatric physical therapy. Evaluation of the videos taught us that physical therapists often do other things than they intended to do (Blauw-Hospers et al 2010). This means that only the evaluation of real action and communication can provide insight into the reality of practice.

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CHAPTER 7

PEDIATRIC PHYSICAL THERAPY IN INFANCY: FROM NIGHTMARE TO DREAM? A RANDOMIZED CONTROLLED TRIAL

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Abstract

Purpose. To study the effects of a new pediatric physiotherapeutic intervention program on the development of infants at high risk for developmental disorders. We used a combined approach in a randomized controlled trial (RCT) and process evaluation.

Subjects and Methods. Forty-six high-risk infants were randomly assigned to receive COPCA (n=21) or traditional infant physical therapy (TIP; n=25) between three to six months corrected age (CA). Developmental outcome was assessed at six and 18 months CA with a neurological examination, AIMS, PEDI, and the mental developmental index (MDI) of the BSID. Contents of the intervention were analyzed by a quantitative video analysis of therapy sessions. Multivariate statistics were applied to evaluate associations between intervention and developmental outcome components.

Results. The RCT revealed that developmental outcome in both groups was largely identical, except for cognitive outcome. The COPCA group decreased less in MDI score from six to 18 months CA (106.5 to 100; p=0.07) than the TIP group (115.5 to 98; p=0.001). Process evaluation showed that typical COPCA actions – 1) family involvement and educational actions, 2) application of a wide variation in challenging the infant to produce motor behavior by him/herself and allowing the infant to continue this activity, and 3) stimulation of motor behavior at the limit of the infant's capabilities – were associated with a better developmental outcome at 18 months CA. The use of handling techniques was associated with a lower score on the PEDI.

Conclusion. Extending the RCT by using process evaluation was needed to obtain insight into associations between the components of intervention and developmental outcome. Specific components of COPCA were associated with improved outcome, while handling techniques were associated with worsened functioning.

Introduction

Research has shown that evidence for the effectiveness of pediatric physical therapy on motor and cognitive development of infants with or at high risk for developmental disorders is inconclusive (Blauw-Hospers and Hadders-Algra 2005, Blauw-Hospers et al 2007, Spittle et al 2007). However, early pediatric physical therapy is widely advocated and desired in the management of high-risk infants. This inconclusiveness may be related to the specific difficulties inherent in studies on early pediatric physical therapeutic intervention. One important difficulty for these studies is the heterogeneity of the study group. Early detection of infants at risk for a developmental disorder implies that the clinical picture of the problems that an infant will eventually develop has not yet manifested itself. Second, there are many variations in the conditions of families such as social-economic status and family routines. Another, equally important factor that might explain the inconclusiveness of results is related to the intervention itself. Treatment application is often eclectic (Howle 2005, Mayston 2005). This would indicate that a broad heterogeneity in implementing a treatment exists, even within treatments that adhere to the same principles (Blauw-Hospers et al 2010). Pediatric physical therapists tend to include in their treatment those techniques which they perceive as helpful, while leaving other techniques out. Presumably this heterogeneity in treatment practice has been brought about by an evolution in treatment techniques and theoretical assumptions. Finally, the outcome measurements that are chosen to evaluate the effect of an intervention might not be the appropriate ones. Usually instruments are chosen out of habit, as well as for practical reasons, and not on the basis of information regarding test accuracy, utility, and theoretical basis (Ketelaar et al 1998, Heineman and Hadders-Algra 2008).

The best method for controlling the impact of the above-mentioned factors is to evaluate the effect of intervention in a randomized controlled trial (RCT). RCTs are offered as the most objective and unbiased design for evaluating intervention programs (Lachin et al 1988). Random allocation of intervention among the participants ensures that differences in group characteristics that may affect outcome are minimized. The aim is to have the groups compared as similarly as possible except for the precisely defined interventions being examined. As in other research designs, RCTs in pediatric physical therapy also have limitations. RCTs are usually designed to determine the effect of intervention by measuring the change in developmental outcome previous to and after the intervention period. Although this is the most straightforward method for measuring effectiveness, it does not explain the underlying mechanisms that might influence outcome (Watson et al 2004). Rarely are the contents of pediatric physical therapy programs analyzed despite the fact that this might serve as an eye-opener in interpreting the results of effectiveness studies. Unraveling the components of intervention shows whether the applied intervention is eclectic or not. Lettinga et al. (2002) stressed the importance of an in-depth understanding of the characteristics of interventions. They stated that every trial which compares the effects of different types of intervention should start with a detailed analysis of the similarities and differences in the content of the interventions under study. This analysis will serve as an addition to discovering the effective and ineffective elements in the interventions, and will consequently result in knowledge about the

implementation of the intervention by different pediatric physical therapists in different settings. A second limitation that an RCT has is related to the validity of using experimental methods in research addressing human activity, which is, for example, the case in early intervention programs. Watson et al. (2004) argue that changes in behavior and psychosocial issues are better evaluated with qualitative approaches, even though these approaches may be less robust.

This paper will describe the results of an RCT on pediatric physical therapy in infancy. In an early intervention project, the VIP project (VIP is the Dutch abbreviation for "Vroege Interventie Project"), we studied the effects of early pediatric physical therapeutic intervention on the developmental outcome of infants with or at high risk for developmental disorders. The effects of a recently developed early intervention program, COPCA (COPing with and CAring for infants with special needs: a family-centered program; Dirks and Hadders-Algra 2003, Dirks et al, submitted for publication), were compared to the effects of traditional infant physical therapy (TIP). To overcome the aforementioned limitations of RCTs, we extended our RCT with a process evaluation to examine how COPCA and TIP were implemented in daily pediatric physical therapeutic practice. Based on the contents of both interventions, we attempted to identify whether changes in developmental outcome were associated with the contents of the intervention or whether they could be explained by other factors. To document developmental changes, a set of instruments was used, ranging from instruments measuring at impairment level (neurological examination) to instruments at the level of activity and participation (PEDI).

Method

Participants

The study groups of the VIP project consisted of infants who had been admitted to the neonatal intensive care unit of the University Medical Center Groningen between March 2003 and May 2005. At ten weeks corrected age, a video recording was made of the infants' General Movements (GMs). Infants and their families were selected to participate in the project when the quality of the infants' GMs was classified as definitely abnormal. The presence of definitely abnormal GMs at fidgety GM age (i.e., at 2-4 months post-term) indicates a high risk for developmental disabilities, such as cerebral palsy (CP) (Prechtl 2001, Hadders-Algra 2004). Infants with severe congenital anomalies and infants whose caregivers had an inappropriate understanding of the Dutch language were excluded from the study. Informed consent was obtained and the research project was approved by the Ethics Committee of the University Medical Center Groningen. The trial is registered under ISRCTN85728836.

Forty-six infants participated in the VIP project. Through block randomization (full-term infants blocks of n=2; preterm infants blocks of n=12) the infants were assigned to COPCA (n=21) or TIP (n=25). The groups were comparable on baseline characteristics, such as gender, gestational age, birth weight, and presence and severity of brain lesions (Table 7.1), but the groups differed for maternal education, which was significantly better in the TIP group (Table 7.1). The randomized intervention was provided between three and six months corrected age (CA). After the intervention period, pediatric physical therapy was only continued when the pediatrician in charge of the infant considered it necessary.

Table 7.1. Baseline characteristics.

		COPCA (n=21)	TIP (n=25)
Gender	n (%)		
▪ Male		9 (43)	11 (44)
▪ Female		12 (57)	14 (56)
Gestational age (wk)	median (range)	29 (27-40)	30 (25-39)
Birth weight (g)	median (range)	1210 (585-4750)	1143 (635-3460)
Maternal age	mean ± SD	30.5 ± 6.2	31.8 ± 4.3
Firstborn child	n (%)	12 (57)	13 (52)
Twin pairs	n (%)	9 (43)	7 (28)
Abnormal cerebral ultrasound	n (%)		
▪ IVH gr. 3-4		1 (5)	1 (4)
▪ PVL gr. 3-4		2 (10)	1 (4)
Maternal education *	n (%)		
▪ Low		3 (14)	3 (12)
▪ Middle		16 (76)	11 (44)
▪ High		2 (10)	11 (44)

* $p<0.05$

SD = standard deviation; wk = weeks; g = gram; gr = grade; IVH, grading according to Volpe (2000) = intraventricular haemorrage; PVL, grading according to de Vries et al (1992) = Periventricular leukomalacia; Levels of education: low = primary education/ junior vocational training, middle = secondary education / senior vocational training, high = university education / vocational colleges

Intervention

TIP

TIP consisted of infant physical therapy as it is applied nowadays in the Netherlands. For the most part it consists of the implementation of the "living concept" of neurodevelopmental treatment (NDT) principles which primarily focus on the sensorimotor functional problems of the infant (Howle 2002, Dirks et al, submitted for publication). More recently, influences from a more functional approach, such as the encouragement of self-produced motor behavior, have also been incorporated into the treatment. The functional approach emphasizes that critical parts of motor learning are finding solutions for new task demands and adaptation to changes in the environmental context (Ketelaar et al 2001, Ekström et al 2005). Due to the different influences which

have been incorporated into TIP, a broad heterogeneity in treatment applications was expected. In the TIP group, intervention was applied with a median value of once a week, mainly in the home environment by the pediatric physical therapist working in the area.

COPCA

COPCA (COPing with and CAring for infants with special needs: a family-centered program) (Dirks and Hadders-Algra 2003, Dirks et al, submitted for publication) was developed from the notion that evidence for the effectiveness of the current pediatric physical therapeutic intervention programs was inconsistent. COPCA differs from existing approaches, both in theoretical background and in implementation (Dirks et al, submitted for publication). It is a family-relationship-oriented program consisting of two theoretical components. The first is formed by a family-involvement and educational-parenting component. A fundamental idea of COPCA is the transactional perspective on family function, in other words, on patterns of reciprocal contingent interactions between caregiver and infant, with the notion that changing patterns of behavior are a characteristic of early development (Fiese 1997, De Weerth and Van Geert 2002). This means that development is seen as a result of a continuous dynamic interplay between child behavior and caregiver responses to the child's behavior, along with environmental variables that may influence both child and caregiver. Caregivers are coached to recognize the infant's signals and to respond appropriately to the actual needs of the infant (Dirks et al, submitted for publication). Coaching implies that the pediatric physical therapist – in the COPCA program, the coach – supports all family members, including the infant with special needs, in order to reveal their competencies, goals, desires, and hopes. On the basis of an ongoing equal partnership in which the family defines the priorities for intervention (Law et al 1998), the coach supports the family in developing their own ways of caring for the infant and in improving personal coping skills. Specific attention is paid to educational actions such as spending brief amounts of time playing in child-preferred activities whereby the motor principles of NGST are taken into account, as well as educational actions promoting appropriate behavior (Dirks et al, submitted for publication).

The second, equally important, theoretical component of COPCA is a neurodevelopmental component based on the principles of Neuronal Group Selection Theory (NGST; Edelman 1993). NGST emphasizes that development is the consequence of a complex interaction between genetic information and environmental influences. According to NGST, development is characterized by two phases of variability, that is, primary and secondary variability. During primary variability the child explores all the variations of motor possibilities that are available in the nervous system. In this phase the child is not yet able to adjust his behavior to external conditions. In typically developing infants, this phase is characterized by abundant variation. At function-specific ages the infant reaches the phase of secondary variability, that is, the child gradually learns to select the most efficient solution for a given task out of his motor repertoire. This selection is based on self-produced trial and error experiences (Hadders-Algra 2000a). Infants with a prenatally, perinatally, or early postnatally acquired lesion or malformation of the brain have a reduced repertoire of motor strategies available for exploration. This is already expressed during the initial postnatal months by a limited repertoire of general movements and continues when goal-directed motility emerges (Hadders-Algra 2000b, 2008a). In addition, these infants have problems with the selection of the most appropriate solution for a certain task out of the repertoire due to deficits in the processing of sensory information (Hadders-Algra 2000b, 2008a). The aim of COPCA is therefore to promote self-produced motor behavior (hands-off), variation, and trial-and-error experiences by means of play, all with the ultimate goal of providing an infant with a wider diversity in terms of neuronal networks that will help the infant to find an appropriate solution for any given motor task.

COPCA was applied twice a week for one hour at home by one of four specially trained pediatric physical therapists.

Developmental outcome

The developmental status of the infants was assessed at baseline three months CA and thereafter at six and 18 months CA. At three and six months, the neurological condition was measured with the Touwen Infant Neurological Examination (TINE; Hadders-Algra et al 2010). The neurological condition was summarized as normal, normal-suboptimal, minor neurological dysfunction (MND), or as abnormal. The classification of abnormal during early infancy implies the presence of a distinct neurological syndrome such as a clear hypo- or hypertonia, a hemi syndrome, or a hyperexcitability syndrome. To distinguish between MND, normal-suboptimal, and normal neurological conditions, the findings of the TINE were classified according to age-specific norms into five clusters of dysfunction: dysfunctional reaching and grasping, dysfunctional gross motor function, brain stem dysfunction, visuomotor dysfunction, and sensorimotor dysfunction. Two forms of normal neurological development could be distinguished: neurologically normal when none of the clusters met the criteria for dysfunction and normal-suboptimal when one or two clusters fulfilled the criteria for dysfunction. When more than two clusters fulfilled the criteria for dysfunction, infants were classified as MND. A recent study has shown that MND can be assessed reliably with the TINE (Hadders-Algra et al 2010).

At 18 months CA, a neurological assessment was carried out according to Hempel (1993). The findings of the Hempel assessment were classified as neurologically normal, simple MND, complex MND, or CP. The distinction between simple and complex MND was also based on the number of clusters of dysfunction. Note that the clusters of dysfunction of the Hempel assessment are similar to that of TINE but not identical. The criteria for classification also differed for both assessments. Simple MND at 18 months denotes the presence of one cluster of dysfunction and complex MND the presence of more than one cluster of dysfunction. The classification CP implies the presence of a "classical" configuration of neurological signs (Hadders-Algra 2003). Little information was available about the concurrent and predictive validity of the Hempel assessment, but the interrater reliability was satisfactory (Hadders-Algra et al 2010).

Additionally, we used the Neurological Optimality Score (NOS; Huisman et al 1995) to summarize neurological condition at 18 months CA. The NOS is the sum of 57 items representing the

neurological examination that meets predefined criteria for optimality. It is important to realize that the definition of optimal is narrower than that of normal or typical, and that a reduced optimality does not always mean abnormal (Prechtl 1980). The NOS has been proven to be an excellent instrument for evaluating subtle differences in neurodevelopmental outcome (Bouwstra et al 2006).

Gross motor development was assessed with the Alberta Infant Motor Scale (AIMS; Piper and Darrah 1994). The AIMS consists of 58 items that evaluate gross motor function in supine, prone, sitting, and standing positions. Through observation of spontaneous motor behavior, each item can be scored on three aspects of motor performance: weight bearing, posture, and antigravity movements. The AIMS has very good reliability coefficients (Piper and Darrah 1994, Heineman and Hadders-Algra 2008). In addition, the validity of the AIMS has been thoroughly examined and proven to be satisfactory (Piper and Darrah 1994, Darrah et al 1998, Liao and Campbell 2004).

The cognitive outcome of the infants was assessed at six and 18 months CA using the mental scale of the Dutch version of the Bayley Scales of Infant Development (BSID-II; Bayley 1993, Van der Meulen et al 2002). The mental developmental index (MDI) consists of items concerning problem solving, memory, discrimination, classification, language, and social skills. Raw scores were converted into age-equivalent scores, as derived from the Dutch norms (Van der Meulen et al 2002). The interrater reliability of the MDI was sufficient; the construct and concurrent validity were moderate (Heineman and Hadders-Algra 2008).

The Pediatric Evaluation of Disability Inventory (PEDI; Haley et al 1992) was used to measure the functional ability of the child. The PEDI was developed for young children from six months to 7.5 years of age and adapted to a Dutch version by Custers et al. (2000a) The PEDI is a discriminative measurement which aims to detect whether a child has limitations in his functional status and, if so, to determine the extent and content area of the limitations. With the PEDI, both the capability of the child (what a child can do) and the performance (what the child actually does) of routine daily childhood activities can be evaluated. Capability is measured by the functional skills scale, while the caregiver assistance scale and the modifications scale provide information on the performance. Each scale consists of three domains, namely, self-care, mobility, and social function. The PEDI can be considered a reliable and valid instrument (Custers et al 2002a, 2002b, Wassenberg-Severijnen et al 2003).

Analysis of the contents of COPCA and TIP

At four and six months CA, we made video recordings of an intervention session. For 43 infants, video recordings of infant physical therapy sessions were available. The remaining three infants (all TIP) had too few intervention sessions to make a video recording. The contents of the sessions were analyzed with a standardized observation protocol that we recently developed for analysis of physiotherapeutic intervention sessions in young infants (Blauw-Hospers et al 2010). The protocol is based on knowledge about infant physical therapy and analysis of directly observable physiotherapeutic (PT) and caregiver actions. Inter- and intrarater agreement proved to be satisfactory

(Blauw-Hospers et al 2010). The observation protocol classifies PT actions into eight main categories: 1) Family involvement and educational actions; 2) Communication; 3) Facilitation; 4) Sensory experience; 5) Passive motor experience; 6) Self-produced motor behavior with no interference; 7) Challenge to self-produce motor behavior where the infant is allowed and challenged to continue activity; 8) Challenge to self-produce motor behavior, flowing over into therapeutic handling. We added a variable which was the amount of postural support provided during PT actions.

The analysis was carried out with the help of the computer program "The Observer" (Noldus). The Observer software is a tool for collecting and analyzing observational data in a descriptive and quantitative way. The outcome parameters of the analysis were the relative amounts of time spent on PT actions. The category self-produced motor behavior without interference of the physical therapist or the caregiver that is present in the original protocol was left out of the current analyses because of a qualitative difference between the two groups during the periods of entirely spontaneous activity (Dirks et al, submitted for publication). Previously, we reported that PT actions at four months were largely comparable to those at six months (Dirks et al, submitted for publication). Assuming that the two measurements at four and six months represented the actions during the intervention period better than a single measurement, we used the average of the four-month and six-month values for PT actions in the correlations with developmental outcome.

Data-analysis

Statistical analyses were performed using the computer package SPSS (version 14.0). To evaluate the effect of type of intervention (COPCA or TIP; group classification on the basis of the video contents of the physical therapy session) on developmental outcome at 6 and 18 months CA, the non-parametric Mann-Whitney test was used since the data were not normally distributed. Differences having a p-value <0.05 were considered statistically significant.

To examine associations between the contents of intervention and developmental outcome, univariate analyses were applied first. To minimize the possibility that associations could be explained by the influence of potential confounders, such as the infant's baseline condition (neurological outcome and AIMS score at three months, the presence of a severe brain lesion), social factors (maternal education, being the first child or not), and factors related to the intensity of physical therapy treatment (number and duration of sessions), we applied multivariate statistics. Linear regression techniques were used, since the data were skewed to a minimal extent only. In this way the contribution of the relative duration of PT actions to outcomes on the NOS, AIMS, BSID-II, and PEDI was assessed. For the associations between the contents of the intervention and the AIMS score at 18 months CA, logistic regression analysis was carried out, given that at 18 months the AIMS suffered from a ceiling effect. Therefore, we dichotomized the AIMS score into 58 and <58. Correlations with a p-value ≤ 0.01 were considered statistically significant.

Results

Developmental outcome according to randomized group allocation

The developmental outcome of the three infants in the TIP group who had too few intervention sessions to make a video recording was similar to the other 22 infants who received TIP. Therefore, we decided to include them in the RCT analysis.

The neurological condition at 6 and 18 months CA in the COPCA and TIP groups was similar (Table 7.2). At six months, one infant in the COPCA group had a normal-suboptimal neurological condition, 16 were classified as MND, and 4 showed an abnormal neurological condition. In the TIP group, 20 infants were classified as MND and 5 had an abnormal neurological condition. At 18 months CA, ten infants, five in each group, had developed CP. One infant in the COPCA group and two infants in the TIP group were neurologically normal. The remaining infants (15 COPCA, 16 TIP) had complex MND. Two infants in the TIP group did not return to the follow-up at 18 months. The NOS of the COPCA group at 18 months tended to be a bit better than the TIP group (COPCA median value 31 versus TIP median value 27: Table 7.2) but the difference did not reach statistical significance.

Performance on the AIMS at 6 and 18 months CA was identical for both groups. This was true for the total AIMS scores and the scores on the subscales (Table 7.2). The data revealed that at the age of 18 months the AIMS suffered from a ceiling effect (Darrah et al 1998, Liao and Campbell 2004). It only could differentiate between children with and without CP.

The MDI scores at 6 and 18 months CA did not differ for COPCA and TIP. At six months CA, the median value of the MDI in the COPCA group was 106.5 and in the TIP group 115.5; at 18 months CA, the median values of the MDI were 100 and 98, respectively. The data also indicated that in both groups the MDI decreased between 6 and 18 months CA. The decrease in MDI score from 6 to 18 months CA was statistically significant in the TIP group (p=0.001; Figure 7.1) but non-significant in the COPCA group (p=0.07). The change in MDI score between 6 and 18 months CA was affected by the level of maternal education (p=0.01) and the type of intervention (p=0.03). The data indicated that especially those infants in the COPCA group whose mother had a lower level of education showed the smallest drop in MDI score (Figure 7.2). At 18 months, there were no differences between COPCA and TIP on the PEDI (Table 7.2).

Table 7.2. Developmental outcome on group level (randomized groups).

6 months CA	**COPCA (n=21)**	**TIP (n=25)**
TINE (n=46)		
Normal-suboptimal	1	0
MND	16	20
Abnormal	4	5
AIMS (median value; range)	18 (6-22)	17 (9-22)
BSID-MDI (median value; range)	106.5 (50-135)	115.5 (84-145)

18 months CA	**COPCA (n=21)**	**TIP (n=23)**
Hempel (n=44)		
Normal	1	2
Complex MND	15	16
Abnormal / CP	5	5
NOS (median value; range)	31 (9-51)	27 (10-47)
AIMS (median value; range)	57 (10-58)	58 (16-58)
BSID-MDI (median value; range)	100 (50-119)	98 (72-135)
PEDI – Functional skills scale (median value; range)		
Selfcare	24 (13-38)	24 (11-27)
Mobility	35 (6-44)	29 (6-37)
Social	21 (11-36)	22 (11-31)

AIMS = Alberta Infant Motor Scale, BSID-MDI = Bayley Scales of Infant Development - mental developmental index, CA = corrected age, CP = cerebral palsy, MND = minor neurological dysfunction, NOS = neurological optimality score, PEDI = Pediatric Evaluation of Disability Inventory, TINE = Touwen Infant Neurological Examination

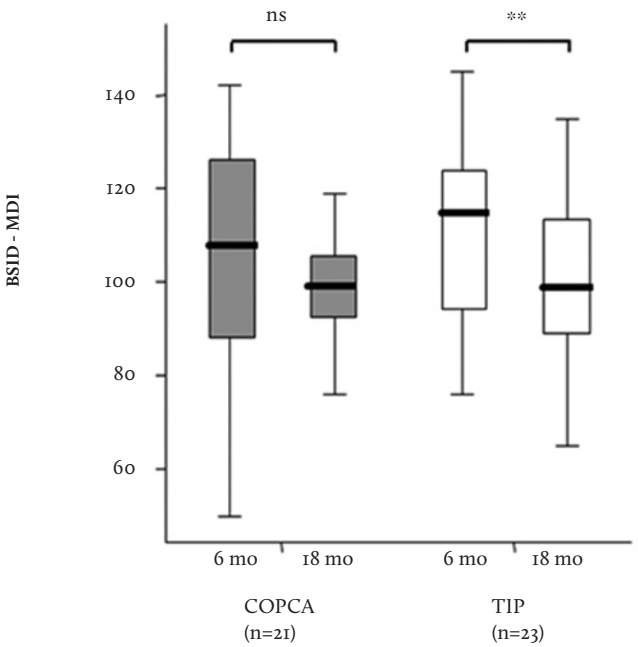

Figure 1: Bayley Scales of Infant Development: MDI scores at 6 and 18 months in the COPCA and TIP group.
Vertical bars, range; horizontal lines, median values; boxes, interquartile range; ns, not significant.
** p=0.001 (Wilcoxon signed ranks test).

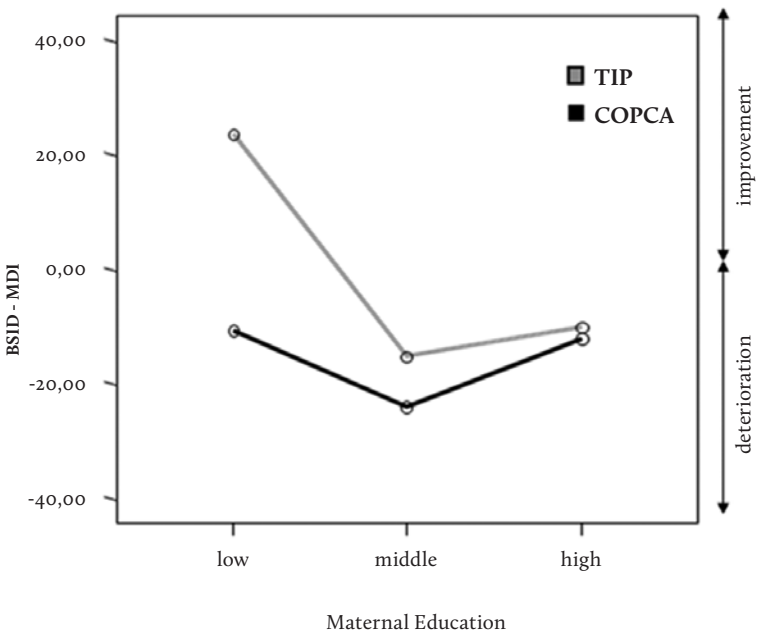

Figure 2: Relative deterioration in MDI score between 6 and 18 months and maternal education. The graphs illustrate that MDI is affected by maternal education (p=0.01) and type of intervention (p=0.03) (MANOVA).

Associations between treatment principles and developmental outcome

The results of the RCT showed only minimal differences between COPCA and TIP. We did, however, observe changes in developmental outcome. To examine whether these changes could be associated with treatment principles used by the physical therapist or caregiver during intervention or with confounding factors, we applied process evaluation to the data. First, we analyzed the contents of the intervention sessions. The details of the analysis are described in Dirks et al. (submitted for publication). Typical COPCA components were: 1) physical therapist coaches the parent (coaching model); 2) to stimulate self-produced motor behavior at the limit of an infant's capabilities; and 3) infant is challenged to produce motor behavior by him/herself and is allowed to continue activity. Typical TIP components were: 1) physical therapist teaches the infant and trains the parent (teacher-learner model); 2) in handling techniques; 3) in sensory stimulation; and 4) to challenge for self-produced motor behavior that is followed by a handling technique.

Preliminary analyses showed that the associations between PT actions and developmental outcome measurements differed for infants with and without CP. An overview of the associations between treatment principles and developmental outcome is presented in Table 7.3.

In infants who developed CP (n=10) no statistically significant associations were found between specific PT actions and developmental outcome at six months CA. Some PT actions were correlated with outcome at 18 months in the infants with CP. The multivariate analyses showed that more time spent on family involvement and educational actions, more time spent with postural support at the limit of the infant's capabilities, and more time during which the infant was challenged in a widely varying way to produce motor behavior and was allowed to continue this activity were associated with a higher score on the functional mobility scale of the PEDI at 18 months CA (all $p<0.01$; Table 7.3). More time spent with passive experience was associated with a lower NOS at 18 months CA ($p<0.01$).

In the children without CP (n=33), the amount of time spent on sensory and passive experience showed a positive correlation with the MDI at six months (both $p<0.01$), but these associations did not persist through to the age of 18 months. In children without CP, more time spent on handling techniques was associated with a lower score on the functional mobility scale of the PEDI at 18 months ($p<0.01$).

Allocation of intervention based on video analysis

Since the treatment application was very heterogeneous, we decided to regroup the infants on the basis of the contents of the video into intervention subgroups. The infants who received COPCA were allocated based on the contents of the intervention to COPCA++ or COPCA+. The intervention sessions of infants who got TIP were classified as TIP++ or TIP+. The ++ notation indicates that the intervention was performed fully according to the principles of COPCA or TIP. In the case of TIP, this meant that the principles of the original concept of the Bobaths were used. The + notation indicated that the contents of the intervention was more diverse. For COPCA this implied that COPCA principles were applied incompletely, while for TIP, that treatment consisted

Table 7.3. Overview of associations between PT-actions and developmental outcome at 6 and 18 months. Results of univariate and multivariate analysis.

a) Infants who developed CP (n=10)

	Outcome measure	Model	Beta	CI	p
Family involvement and educational actions					
U	PEDI - functional mobility scale 18 mo	Fameduc	0.54	7.59-70.24	0.02
M	PEDI - functional mobility scale 18 mo	Fameduc AIMS 3	0.52 0.51	11.91-63.66 0.60-3.40	<0.01 <0.01
Postural support - at the verge of infant's capabilities					
U	PEDI - functional mobility scale 18 mo	Support verge	0.60	12.85-69.59	<0.01
M	PEDI - functional mobility scale 18 mo	Support verge AIMS 3	0.56 0.48	15.09-62.24 0.56-3.24	<0.01 <0.01
Infant challenged to produce motor behavior and allowed to continue activity (large variation offered)					
U	PEDI - functional mobility scale 18 mo	CPMB - allowed large var	0.52	4.37-47.72	0.02
M	PEDI - functional mobility scale 18 mo	CPMB - allowed large var AIMS 3	0.55 0.56	10.41-44.57 0.83-3.53	<0.01 <0.01
Passive motor experience					
U	NOS 18 mo	Passive	-0.61	-112.69- -22.91	<0.01
M	NOS 18 mo	Passive Frequency prior to 3 mo	-0.79 0.46	-92.00- -54.72 0.05- -0.12	<0.01 <0.01

b) Infants without CP (n=33)

	Outcome measure	Model	Beta	CI	p
Sensory experience					
U	MDI 6	Sensory	0.34	19.88-155.07	0.01
M	MDI 6	Sensory AIMS 3	0.36 0.32	28.36-157.09 1.00-7.79	<0.01 0.01
Passive motor experience					
U	MDI 6	Passive	0.29	11.69-218.80	0.03
M	MDI 6	Passive Neur exam 3 mo	0.35 -0.34	38.77-237.60 -26.47- -3.91	<0.01 <0.01
Facilitation techniques					
U	PEDI - functional mobility scale 18 mo	Facilitation	-0.41	-23.86- -6.00	<0.01
M	PEDI - functional mobility scale 18 mo	Facilitation AIMS 3 mo	-0.36 0.25	-22.03- -4.29 0.02-2.69	<0.01 0.05

AIMS = Alberta Infant Motor Scale, CPMB - allowed large var = Infant challenged to produce motor behavior and allowed to continue activity (large variation offered), Fameduc = Family involvement and educational actions, Frequency = number of treatment sessions, MDI = Mental Developmental Index, M = multivariate, Neur exam = neurological examination, NOS = neurological optimality score, PEDI = Pediatric Evaluation of Disability Inventory, U = univariate

of a mix of actions according to the original Bobath concepts along with current NDT principles involving a more functional approach.

The classification COPCA++ versus COPCA+ was based upon the amount of time that was spent on the typical COPCA actions where the physical therapist coaches the caregiver to stimulate motor behavior at the limit of an infant's capabilities and where the infant is challenged to self-produce motor behavior that is then continued by the infant. The distinction TIP+ versus TIP++ was made based on the time spent on training the caregiver, handling and pressure techniques, sensory experience, a challenge to self-produce motor behavior followed by a handling technique, and the amount of time that actions were performed while the pelvis of the infant in supine position was lifted slightly by the hands of the physical therapist. Each infant was given a score on the basis of the average time-values of PT actions observed during the two intervention sessions on the basis of the percentile values of the present study. When the time spent on a PT action was below the 33rd percentile, 1 point was given; a time score between the 33rd and 67th percentile resulted in 2 points; and a time score over the 67th percentile was given 3 points. In this way a total COPCA score (maximum: 3 actions * 3 points = 9) and a total TIP score (maximum: 5 actions * 3 points = 15) were calculated. The cut-off for the distinction between the ++ and + classification was the 67th percentile of the total score. This resulted in a new group allocation that is presented in Table 7.4. The allocation of the groups based on the time spent on PT actions matched the classification of the second author (TD) on the basis of Gestalt perception of the video recording.

Table 7.4. Reallocation into intervention groups.

Classification	Total group (n=43)	Infants with CP (n=10)	Infants without CP (n=32)*
TIP ++	14	3	11
TIP +	8	2	5
COPCA +	7	1	6
COPCA ++	14	4	10

*1 infant did not return to the follow up.
TIP++ = classic NDT approach
TIP+ = mix of classic NDT and a more functional approach
COPCA+ = COPCA principles applied incompletely
COPCA++ = COPCA principles fully applied

Based on the new group allocation, we reanalyzed the outcome measurements. The groups did not differ in neurological classification, and AIMS and MDI scores at 6 and 18 months. For the domain of mobility of the functional skills scale of the PEDI at 18 months, the infants who received COPCA or TIP+ performed significantly better than the infants in the TIP++ group (Mann-Whitney $p < 0.01$; Figure 7.3a). They also needed less caregiver assistance in the mobility domain at 18 months ($p=0.03$; Figure 7.3b). The differences in performance for both PEDI outcomes between children who received COPCA or TIP+ and those who received TIP++ remained statistically significant in the multivariate analyses (details not provided).

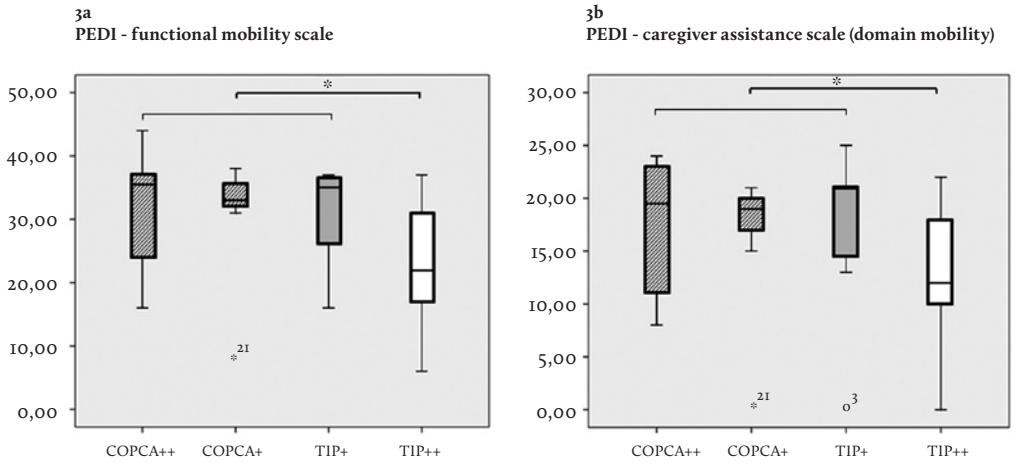

Figure 7.3a: Scores on the domain mobility of the functional skills scale of the PEDI at 18 months CA in the COPCA+, COPCA++, TIP+ and TIP++ groups.
Vertical bars, range; horizontal lines, median values; boxes, interquartile range.
* difference between TIP++ vs other three groups: $p < 0.01$, a significance which persisted in the multivariate analysis.

Figure 7.3b: Scores on the caregiver assistance scale in the domain mobility of the PEDI at 18 months in the COPCA+, COPCA++, TIP+ and TIP++ groups.
Vertical bars, range; horizontal lines, median values; boxes, interquartile range.
**difference between TIP++ vs other three groups: $p=0.03$

Discussion

The RCT revealed that COPCA and TIP were associated with similar effects for developmental outcome. But the process evaluation indicated that the virtual absence of difference in group effect may partially have been caused by the extensive heterogeneity in intervention strategies within the groups, especially within the TIP group. The process analysis indicated that there was some evidence that important components of the COPCA intervention were associated with an improved developmental outcome. The therapeutic components of intervention during early infancy, which were associated with a better developmental outcome at 18 months, were family involvement and educational actions, application of broad variations in challenging the infant to produce motor behavior by him/herself and allowing the infant to continue this activity, and the provision of postural support at the limit of an infant's capabilities.

Methodological considerations

The major strength of this study was its double approach. The combination of the RCT with a process evaluation made it possible to determine the influence of the intervention on developmental outcome. Another strength of this study was that the infants all received intervention during the same age period, that is, from three to six months post-term. This made the COPCA and TIP groups more comparable. There was hardly any attrition during the study. Only two infants did not return to the follow-up at 18 months CA. The final strength was that we used a set of instruments to document outcome, ranging from instruments measuring at impairment level (neurological examination) to instruments measuring the level of activity and participation (PEDI).

One of the limitations of this study was that we studied a relatively small group of 46 infants. Although the infants were selected because they all showed definitely abnormal GMs at ten weeks CA, they had a heterogeneous developmental outcome. Various studies indicated that the presence of definitely abnormal GM is associated with a high risk for developmental motor disorders such as cerebral palsy (Prechtl 2001, Hadders-Algra 2004). In this study only a minority of the infants was diagnosed with CP at 18 months. This limitation is inherent to developmental changes in the central nervous system. Developmental outcome at 18 months CA was relatively good, since the median values of the MDI were 98-100. It should be noted that the majority of infants showed complex MND at 18 months, which puts the infant at risk for learning and behavioral disorders at school age (Hadders-Algra 2005). Another limitation is that we used 18 months CA in this study to evaluate the long-term outcome of the intervention. As mentioned above, some developmental problems do not emerge before school age. Therefore, we recommend that future research should re-examine these children at school age. Finally, the major limitation of this study was the heterogeneity of the composition of treatment in the TIP group. We anticipated this problem by including a process analysis, thereby turning a major limitation into a major strength.

Physiotherapeutic considerations

The RCT revealed only minor differences between the COPCA and the TIP group. One possible explanation for the small size of the effect is that intervention can affect developmental outcome in children with brain dysfunction to a limited extent only (Ade-Hall and Moore 2000, Wasiak et al 2004, Hoare et al 2007, Anttila et al 2008). Animal and human studies indicate that intervention after a lesion of the brain at early age affects motor development considerably less than cognitive development (Spittle et al 2007, Kolb and Gibb 2007). Another explanation for the lack of effect could be the heterogeneity of the TIP treatment. For research purposes, it is quite nightmarish that treatment application, although based on the same theoretical background, can be so diverse. In our opinion this heterogeneity is a major problem in the application of the RCT design in pediatric physical therapy.

The RCT had two interesting results. First, there was an interaction effect between the type of intervention and the level of maternal education in the relative drop in MDI score between 6 and 18 months CA. Infants whose mothers had a lower level of education showed less decline in MDI score over time than infants whose mothers had completed higher education. This effect was especially present in the COPCA group. This suggests that mothers with lower education especially benefit more from coaching by a therapist who uses COPCA principles, while mothers with higher education are less affected by coaching as well as training performed by a therapist. Second, the finding of a similar neurological outcome in the COPCA and TIP groups is intriguing considering the diametrically opposed view of TIP and COPCA on the importance of the neurological parameters of muscle tone and atypical movements. In the TIP treatment, especially when applied to the original concepts of Bobath and/or NDT, influencing tone by means of handling techniques plays an important role in the activities of the therapist during intervention sessions. In COPCA no attention is paid to these impairments. In other words, handling techniques aimed at influencing muscle tone and facilitating movement sequences to improve function do not seem to affect neurological outcome.

Process evaluation indicated that specific components of the interventions in the study were associated with an improvement in developmental outcome. We would like to stress that observed associations are not causations. Associations between PT actions and outcome are unavoidably contaminated by the child's initial degree of impairment. An infant with a more serious impairment elicits differences in the contents of PT actions than an infant with milder impairments where the degree of initial impairment to a substantial extent determines later outcome. Our data indeed revealed clear interactions between the infant's condition and therapist actions. We therefore studied associations separately for infants with and without CP. In addition, we used multivariate analysis in order to be able to take into account the infant's initial degree of impairment. Nevertheless, it is also conceivable that other infant-specific factors which we did not include in the analyses did affect the choice of PT actions and the child's developmental outcome. Yet, the results of the multivariate analysis may guide our thinking about the effectiveness of specific PT actions.

Several components of COPCA were associated with a higher score in the domain of mobility on the functional skills scale of the PEDI. An example of this is the category of family involvement and educational actions which in COPCA implies coaching of family members to develop their own ways to care for the infant and to cope with the problems of the infant with special needs. During the intervention, the coach listens, informs, and observes (hands-off), while the caregiver is involved in daily routines with the child, including play, thereby creating a situation in which caregivers feel free to explore and discuss alternative strategies. Second, wide variation in self-produced motor activities (hands-off), trial-and-error experiences, and, if necessary, the provision of minimal postural support creates a challenging environment in which the infant may explore and practice his/her motor possibilities (Dirks et al, submitted for publication).

Some typical TIP actions were also associated with developmental outcome. The use of facilitation techniques such as handling, which are applied to reduce atypical functional activities and to prepare the infant for optimally independent function (Dirks et al, submitted for publication), were associated with a worse performance on the functional skills scale of the PEDI. On the other hand, sensory and passive experiences were associated with a higher MDI immediately after the intervention period. Perhaps sensory and passive experiences reflect situations of increased infant attention which may promote cognitive development (Bahrick et al 2004). Our data indicated that this beneficial effect disappeared over time.

Since the treatment application was very heterogeneous, we decided to regroup the infants on the basis of the contents of the video into intervention subgroups. Although this approach lacks the advantages of an RCT, it was interesting to see that infants in the COPCA and functional TIP group scored better for domain of mobility on the functional skills scale and needed less caregiver assistance for domain of mobility on the PEDI than infants who received a TIP treatment according to the original Bobath approach. This would indicate that the functionally oriented interventions succeeded in influencing the performance of functional skills in a beneficial way. It was interesting to observe that the effects of intervention were clearer at 18 months CA than immediately after the intervention at 6 months CA. Perhaps this finding is related to the COPCA approach which aims at supporting family members on the basis of an ongoing equal partnership in order to uncover their own specific problem-solving strategies for caring and coping with the infant with special needs.

Concluding remarks

The study showed that our nightmare had a good outcome. Extending the RCT with process evaluation was necessary in order to answer the question of what components of intervention are associated with developmental outcome. Our study showed that specific components of COPCA, especially family involvement and educational actions, challenging the infant with a wide variation to produce motor behavior by him/herself and then to allow the infant to continue this activity, and stimulating motor behavior at the limit of an infant's capabilities according to the principles of the NGST, were associated with improved developmental outcome.

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CHAPTER 8

ADDITIONAL RESULTS ON OUTCOME MEASURES OF THE VIP PROJECT

In this Chapter some of the results of questionnaires which were also assessed during the VIP project, but were not the focus in the previous sections, are presented. Successively, attention is paid to the Kent Infant Development Scales (KIDS), the Measures of Processes of Care (MPOC) and the Nijmegen Questionnaire on Child-rearing Situations (NCSQ). Furthermore some preliminary results on postural control will be described. The results of the IMP, video recordings of ADL activities and postural control will be published separately.

Kent Infant Development Scales

In total, 113 questionnaires (51 COPCA; 62 TIP) were returned (response rate of 82%). Especially at 3 months CA attrition was high (28%). At 6 and 18 months CA attrition was lower, but still ranged from 8 to 19%. One possible explanation for the attrition rate was that despite the instructions some caregivers did not complete the questionnaire before the next test session. Reasons they mentioned were that they did not exactly know the corrected age of the infant, they didn't see the necessity to fill out the questionnaire at the right time, or they simply forgot. Caregivers with a lower educational level or with known psychopathological problems tended to return the questionnaires less often.

The results showed that KIDS scores of infants in the COPCA group did not differ from those in the TIP group. Both the total score and the scores on the five subscales were comparable at 3, 6, and 18 months CA (see Table 8.1). The possibility that under- or overestimation of the development of the infant influenced the KIDS scores is present but almost negligible. The caregivers were only asked to report whether they had noticed a specific behaviour, not to judge the development of the infant (Stancin et al 1984).Some outliers were present in the data but they hardly influenced the results.

Table 8.1. Median scores and ranges of the KIDS at 3, 6 and 18 months CA.

	COPCA			TIP		
	3 (n=15)	6 (n=17)	18 (n=18)	3 (n=18)	6 (n=23)	18 (n=21)
Total	68.0 (62-122)	139 (106-189)	243.5 (214-252)	64.0 (31-118)	128.0 (77-221)	246.0 (177-252)
Cognition	16.0 (5-25)	33.5 (24-43)	50.5 (44-52)	15.5 (7-28)	31.0 (14-45)	50.0 (42-52)
Motor	16.0 (4-28)	37.5 (24-65)	77.0 (48-78)	15.0 (4-38)	33.0 (21-72)	75.0 (34-78)
Language	11.0 (5-18)	20.0 (9-24)	37.0 (30-38)	10.0 (3-17)	19.0 (11-34)	37.0 (29-38)
Self-help	8.0 (3-20)	23.5 (15-33)	38.0 (29-39)	6.5 (3-11)	21.0 (11-36)	38.0 (29-39)
Social	16.0 (8-27)	29.0 (22-37)	50.0 (44-51)	15.5 (8-25)	26.0 (13-44)	50.0 (38-51)

Measures of processes of Care (MPOC)

In total, 115 questionnaires (53 COPCA; 62 TIP) were returned (response rate of 83%). Caregivers with a lower educational level or with known psychopathological problems tended to return the questionnaires less often. From the 115 questionnaires only 81 (37 COPCA; 46 TIP) were valid and could be used for the final analysis. The non-valid questionnaires contained a considerable amount of missing data, i.e. more than 29 items, resulting in too few answers to calculate a domain score (Van Schie et al 2004). The large amount of non-valid questionnaires was mainly caused by the frequent use of the category 'not applicable'. At both 3 and 6 months CA eleven questionnaires were non-valid. At 18 months CA twelve questionnaires were non-valid. The caregivers of the latter twelve infants reported that they had no experience with a rehabilitation setting or hospital visit during the period between 6 and 18 months CA and judged the MPOC as not suitable for their situation. They did however return the questionnaire, but answered most questions with 'not applicable'.

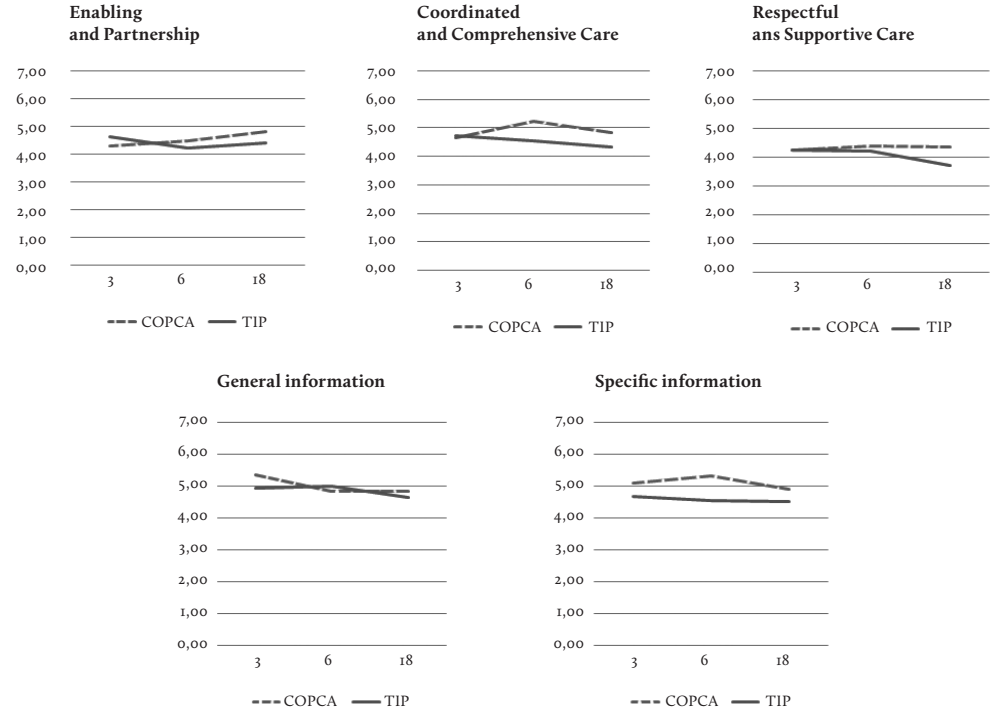

Figure 8.1. Median values of COPCA and TIP on the five domains of the MPOC.

Table 8.2. Median Scores and Ranges of the MPOC at 3, 6 and 18 months CA.

	COPCA				TIP				
	mo	n*	median	Range		mo	n*	median	Range
EP	3	13	4.3	3.9-6.6	EP	3	15	4.6	3.6-5.7
	6	9	4.5	3.7-5.6		6	15	4.2	3.5-4.9
	18	13	4.8	3.9-5.4		18	11	4.4	3.4-5.6
PGI	3	10	5.3	3.7-6.2	PGI	3	15	4.9	3.0-5.9
	6	6	4.8	3.6-6.7		6	13	5.0	2.8-5.7
	18	7	4.8	4.4-5.6		18	8	4.6	3.1-5.2
PSI	3	13	5.1	4.1-6.2	PSI	3	14	4.7	3.7-6.4
	6	8	5.3	3.7-6.0		6	17	4.5	2.9-5.6
	18	11	4.9	3.5-5.7		18	11	4.5	3.5-5.1
CCC	3	13	4.7	4.2-6.7	CCC	3	13	4.7	3.5-5.4
	6	7	5.2	3.9-5.5		6	16	4.6	3.5-5.4
	18	12	4.8	3.7-5.5		18	11	4.3	3.6-6.1
RS	3	12	4.3	3.6-6.4	RS	3	15	4.3	3.2-5.7
	6	10	4.4	3.7-6.1		6	16	4.2	3.6-5.0
	18	14	4.4	3.4-6.3		18	10	3.7	3.2-5.6

CCC = coordinated and comprehensive care for child and family, EP = Enabling and partnership, mo = months corrected age, PGI = providing general information, PSI = providing specific information, RS = respectful and supportive care
*n varies due to missing data on the specific domains

In accordance with the literature (van Schie et al 2004, Nijhuis et al 2007) we noted problems with the MPOC caregiver ratings. Caregivers reported difficulties with judging the care provided by different service providers. Often they had medical experiences with service providers in different settings such as the University Medical Center and a regional hospital. Experience frequently did not match. While participating in the VIP project they could also judge the attitude of the physiotherapist. Therefore we recommend in line with Nijhuis et al (2007) that in future research the family-centredness of different settings should be assessed separately.

Figure 8.1 and Table 8.2 represent the MPOC median scores and ranges. The median scores indicated that caregivers in the COPCA intervention group in most domains are more satisfied with the care they received than caregivers in the TIP group. Furthermore the satisfaction of caregivers in the COPCA group with the care they received improved during the intervention period on the domains enabling and partnership, coordinated and comprehensive care, and respectful and supportive care. Caregivers in the TIP group became less content with the care they received during the intervention period. Note however that none of the findings reached statistical significance.

Nijmegen Child-rearing Situation Questionnaire

In total, 111 questionnaires (52 COPCA, 59 TIP) were returned (response rate of 80%). Thirteen

percent of the caregivers indicated that they refused to fill out the questionnaire because of the confronting nature of some of the questions. They were confronted with questions like "it would have been better when my child had not survived" or "my child should be placed in a fosterhome". As explanation they mentioned the stress they perceived during the NICU stay whether their child would survive or the loss of a twin brother or sister of their child. For this reason the NCSQ probably is not the most suitable instrument to measure parental stress and the existing parenting situation in a study group with high risk infants.

Only a few differences on subscales were observed between COPCA and TIP on the NCSQ (Table 8.3). At 6 months caregivers in the COPCA group are more willingly to accept help regarding educational matters. At 18 months caregivers in the TIP group are more optimistic about the future perspective and they believe that their situation is more dependent on fate.

Postural control

Preliminary results on the first 20 infants of the VIP project showed that at 4 months CA infants of the COPCA (n=9) and the TIP group (n=11) showed less direction-specific activity in supine and sitting than age-matched typically developing infants. At the age of 6 months CA postural control in the group of infants who had received COPCA intervention was in some aspects significantly better than that of the group who had received TIP. Infants who received COPCA showed more direction specific adjustments in sitting, more often recruited the complete pattern in supine and sitting position and showed significantly less often a synchronous onset of postural muscle activity (de Graaf-Peters et al 2007). Postural performance at 6 months of the high risk infants who had received COPCA intervention had improved to such an extent that it closely resembled that of typically developing peers. This indicates that it is conceivable that the postural control of infants at high risk for a developmental motor disorder may be influenced with the COPCA intervention. Whether these results hold for the entire study group and whether they are still present at 18 months CA is currently being assessed.

Summarizing, the additional results show that the scores of the infants in the COPCA and TIP group on the KIDS total score and subscores were comparable at 3, 6 and 18 months CA, findings which are in line with the results which used the Mental Developmental Index of the Bayley Scales of Infant Development as outcome parameter. Outcome in the various domains of the MPOC indicated that there were no statistically significant differences between the COPCA and TIP group at 3, 6 and 18 months CA. However, caregivers in the COPCA group tended to become more satisfied with the care they received during the study while the caregivers in the TIP group became less content. The NCSQ showed some minor differences between the COPCA and TIP group, but probably the questionnaire is not the most suitable instrument to measure parental stress and the existing parenting situation in a study group with high risk infants due to the confronting nature of some of the questions. Finally, preliminary data of the first half of the study group suggested that COPCA presumably has a beneficial effect on postural control.

Table 8.3. Median scores and ranges of the NCSQ at 3, 6 and 18 months CA.

	COPCA			TIP		
	3 (n=17)	6 (n=17)	18 (n=18)	3 (n=19)	6 (n=22)	18 (n=18)
A. Subjective Family Stress	**1.3 (1.0-4.0)**	**1.4 (1.0-4.8)**	**1.3 (1.0-4.3)**	**1.3 (1.0-3.3)**	**1.3 (1.0-4.3)**	**1.3 (1.0-3.8)**
Acceptance	1.0 (1.0-3.0)	1.4 (1.0-3.8)	1.0 (1.0-4.3)	1.0 (1.0-2.3)	1.0 (1.0-2.8)	1.0 (1.0-3.8)
Coping	1.5 (1.0-2.0)	1.5 (1.0-2.9)	1.4 (1.0-2.4)	1.4 (1.0-2.3)	1.5 (1.0-1.9)	1.4 (1.0-2.1)
Problems	1.3 (1.0-2.9)	1.4 (1.0-3.7)	1.5 (1.0-3.1)	1.1 (1.0-3.0)	1.4 (1.0-2.6)	1.7 (1.0-2.9)
Need for change	1.2 (1.0-2.0)	1.2 (1.0-3.2)	1.2 (1.0-2.7)	1.2 (1.0-2.0)	1.2 (1.0-1.7)	1.2 (1.0-1.8)
Strain	1.6 (1.1-3.6)	1.9 (1.0-4.1)	1.6 (1-3.9)	1.7 (1.0-3.3)	1.6 (1.0-2.4)	1.6 (1.0-3.6)
Managing on one's own	1.5 (1.0-4.0)	1.5 (1.0-4.8)	1.5 (1.0-4.0)	1.8 (1.0-2.8)	1.5 (1.0-4.3)	1.5 (1.0-3.8)
Pleasure	1.0 (1.0-1.8)	1.2 (1.0-2.4)	1.0 (1.0-1.8)	1.2 (1.0-1.8)	1.0 (1.0-2.0)	1.1 (1.0-2.0)
Relation	1.4 (1.0-2.0)	1.3 (1.0-3.0)	1.2 (1.0-2.4)	1.4 (1.0-2.4)	1.2 (1.0-2.4)	1.2 (1.0-1.8)
C. Attributions on the childrearing situation	**1.8 (1.0-4.0)**	**2.0 (1.0-4.1)**	**1.8 (1.0-4.7)**	**2.0 (1.0-3.8)**	**1.8 (1.0-4.2)**	**1.8 (1.0-4.2)**
Effort of self and partner	1.8 (1.1-2.9)	2.0 (1.2-2.8)	1.8 (1.1-2.8)	2.0 (1.1-3.1)	1.8 (1.1-3.1)	1.8 (1.1-2.4)
Ability	1.8 (1.0-2.6)	2.0 (1.0-3.2)	1.8 (1.0-2.6)	2.0 (1.0-2.8)	1.8 (1.0-2.6)	1.8 (1.0-2.4)
Share of partner	1.6 (1.2-3.0)	1.9 (1.2-2.6)	1.9 (1.2-2.6)	1.6 (1.0-3.8)	1.8 (1.2-2.8)	1.6 (1.2-3.2)
Task difficulty	1.3 (1.0-3.6)	1.8 (1.0-4.1)	1.2 (1.0-3.8)	1.4 (1.0-2.4)	1.4 (1.0-2.2)	1.6 (1.0-3.1)
Luck/fate	2.2 (1.0-4.0)	2.2 (1.0-3.3)	***1.7**** ***(1.0-4.7)***	2.0 (1.0-3.3)	2.5 (1.0-4.2)	***2.5**** ***(1.0-4.2)***
D. Expectation for help	**1.9 (1.0-5.0)**	**2.3 (1.0-5.0)**	**1.6 (1.0-5.0)**	**2.1 (1.0-5.0)**	**2.0 (1.0-5.0)**	**2.0 (1.0-5.0)**
Satisfaction	4.5 (3.2-5.0)	4.2 (3.7-5.0)	4.5 (3.8-5.0)	4.2 (2.3-5.0)	4.5 (3.5-5.0)	4.3 (3.7-5.0)
Need for change	1.7 (1.0-2.7)	1.9 (1.2-3.3)	1.7 (1.0-3.0)	1.8 (1.0-3.2)	1.8 (1.0-2.5)	1.7 (1.2-2.3)
Need for help	1.4 (1.0-3.0)	***1.8**** ***(1.0-3.0)***	1.0 (1.0-3.2)	2.0 (1.0-3.2)	***1.1**** ***(1.0-2.5)***	1.3 (1.0-2.0)
Expection for help (internal orientation)	2.1 (1.0-3.5)	2.6 (1.3-4.0)	1.6 (1.0-4.3)	2.3 (1.0-4.0)	2.1 (1.0-3.4)	2.1 (1.4-2.8)
Expection for help (external orientation)	1.5 (1.0-3.0)	2.1 (1.0-4.2)	1.0 (1.0-4.0)	2.0 (1.0-2.8)	1.8 (1.0-2.3)	1.8 (1.0-2.3)
Future perspective	2.5 (1.5-3.5)	2.6 (1.0-3.3)	***2.7**** ***(1.0-4.3)***	2.8 (1.0-3.7)	3.0 (2.0-3.7)	***3.3**** ***(1.8-4.3)***

* p<0.05; Mann-Whitney U-test.

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CHAPTER 9

GENERAL DISCUSSION

The main aim of this thesis was to contribute to the body of knowledge about the effects of early physiotherapeutic intervention for young infants who are considered to be at risk for a developmental motor disorder. Therefore existing programmes on the effects of early physiotherapeutic intervention on motor and cognitive development were reviewed and a new physiotherapeutic intervention programme COPCA was evaluated. The evaluation of the COPCA programme not only focused on the effects on developmental outcome but also included a process evaluation of the contents of the COPCA programme. Section 9.1 discusses the implications of the main findings of the studies in this thesis. Reflections about the VIP project are offered in section 9.2. This discussion ends with some implications for physiotherapeutic practice and recommendations for further research (section 9.3).

9.1 Implications of the main findings

9.1.1 The effect of early intervention on developmental outcome

The systematic reviews on the effects of existing early physiotherapeutic intervention programmes on motor (*Chapter 3 and 4*) and cognitive development (*Chapter 4*) of infants with a high risk for developmental motor disorders aged less than 18 months corrected age (CA) resulted in three important findings. First, the effect of early intervention programmes which focus primarily on the improvement of motor development is inconclusive. It remains a matter of debate to what extent early intervention may facilitate outcome in various developmental domains. General developmental programmes seem most promising to influence motor and cognitive development. Second, we recommend that the choice of an early intervention programme should be adapted to the infant's age. It turned out that the type of intervention which might be beneficial for infants at preterm age differs from the type which is effective in infants who have reached at least term age. At preterm age infants seem to benefit most from intervention which aims at mimicking the intrauterine environment, such as NIDCAP intervention. After term age, general developmental programmes in which parents learn how to promote infant development can exert a positive effect on motor and cognitive development. This finding is consistent with the results of studies on the effect of early intervention which primarily aim at improving cognitive outcome. Third, intervention programmes according to the principles of NDT or Vojta do not show a beneficial effect on motor or cognitive development in infants at high risk for developmental disorders in the first postnatal years. A limitation of the systematic reviews was that the contents of the programmes and the methodological quality differed too much to allow for a meta-analysis of the data. The findings of our reviews are in line with similar reviews performed by others investigating the effect of early intervention (Simeonsson et al 1982, Parette and Hourcade 1984, Casto and Mastropieri 1986, Shonkoff and Hauser-Cram 1987, Parry 1992, Turnbull 1993, Guralnick 1997, Majnemer 1998, Butler and Darrah 2001, Spittle et al 2007).

One of the aims of the VIP project was to evaluate the effect of two early intervention programmes on developmental outcome. COPCA can be considered as an example of a general developmental programme while TIP, the traditional approach towards infant physical therapy in the

Netherlands, is based for an important part on the principles of NDT treatment. Overall we found only minor differences in developmental outcome till the age of 18 months between the COPCA and TIP group. A possible explanation for this small size of the effect is that intervention can affect developmental outcome in children with brain dysfunction to a limited extent only (Ade-Hall and Moore 2000, Wasiak et al 2004, Hoare et al 2007, Anttila et al 2008).

Developmental outcome of the infants in the VIP project was relatively good as the median values of the MDI were 98-100 and only a minor part of the groups were diagnosed with CP at the age of 18 months CA. It should be noted however that the majority of infants showed complex MND at 18 months, which puts the infant at risk for learning and behavioural disorders at school age (Hadders-Algra 2005). Important findings of the VIP project were: 1) Neurological outcome at 6 and 18 months in the COPCA and TIP group was similar. This was interesting considering the position that muscle tone and abnormal movement patterns, which are two important aspects of neurological condition, have in both interventions. In TIP normalization of muscle tone and movement patterns plays an important role (Howle 2002), while in COPCA no attention is paid to these impairments. In other words, therapeutic techniques aiming to influence muscle tone and movement patterns, do not seem to affect neurological outcome. 2) Motor performance as measured with the AIMS was similar at 6 and 18 months CA. Where the preliminary analyses for the first half of the study group showed that infants who received COPCA tended to perform better on de sitting subscale of the AIMS (*Chapter 4*), this result could not be replicated when we analyzed the data of the entire study group (*Chapter 7*). 3) Although the overall cognitive outcome was comparable between the COPCA and TIP group at 6 and 18 months CA, we observed a decrease in the MDI score over time which was statistically significant in the TIP group. This decrease in cognitive outcome over time is in line with findings in other studies (Vohr and Garcia Coll 1985, Aylward 1989, Bhutta et al 2002, Ment et al 2003). The change in MDI score between 6 and 18 months CA was affected by the level of maternal education and the type of intervention. Infants whose mother had a lower level of education showed less decline in MDI score over time than infants whose mother had finished a higher education. This effect was in particular present in the COPCA group. This suggests that in particular mothers with a lower education benefit more from the suggestions made by a physical therapist that uses COPCA concepts, while mothers with a higher education seem less affected by suggestions of a physical therapist. 4) At 18 months CA there were no differences between COPCA and TIP on the PEDI. However, it turned out that the score on the functional mobility scale of the PEDI was associated with several elements of the intervention (*see 9.1.2*). 5) Results from the questionnaires (*Chapter 8*) indicated that caregivers in the COPCA group tended to become more satisfied with the care they received during the study while the caregivers in the TIP group became less content (MPOC). Moreover the caregivers in the COPCA group were more willingly to accept help regarding educational matters (NCSQ). 6) Data from the first half of the study group suggested that COPCA presumably has a beneficial effect on postural control. Infants who received COPCA showed more direction specific adjustments in sitting, more often recruited the complete pattern in supine and sitting position and

showed significantly less often a synchronous onset of postural muscle activity (de Graaf-Peters et al 2007).

9.1.2. Evaluation of the application of COPCA in daily practice.

The application of COPCA in daily practice was evaluated by means of video analysis. This process evaluation is a prerequisite for the implementation of COPCA within paediatric physiotherapy. It is acknowledged that designing intervention programmes requires more fundamental knowledge about the relations between function, activities and participation than is offered by RCT research (May et al 2007, Hullegie 2009). Complex therapeutic interventions comprise a number of separate elements which seem essential to the proper functioning of the interventions, which may act both independently and interdependently. The components usually include behaviours, parameters of behaviours (e.g. frequency, timing), and methods of organizing and delivering those behaviours (e.g. type(s) of practitioner, setting and location) (Medical Research Council 2000).

Randomised controlled trials (RCTs) are considered to be the most rigorous way to evaluate the effectiveness of interventions, regardless of their complexity. Because of their multifaceted nature and dependence on social context, complex interventions pose methodological challenges, and require adaptations to the standard design of such trials (Oakley et al 2006). Process evaluation is needed to describe what really happens in an intervention and to explore hypotheses about why and how interventions are effective or ineffective (Lettinga et al 2002, Flottorp et al 2003, Oakley et al 2006). Process evaluation is especially necessary in trials, where the 'same' intervention principles may be implemented, but applied and received in different ways. This thesis contributes to these issues in two ways. First, by developing a standardized observation protocol which allowed for an objective quantification of the contents of physical add therapy in infancy. Second, by combining the RCT with the process evaluation it was actually possible to explore whether specific aspects in the contents of intervention could be associated with developmental outcome. The study described in *Chapter 7* in which we extended the results of the RCT with process evaluation is a good example of integrating the knowledge about the contents of intervention with the scientific approach of a RCT. The process evaluation has helped deepen our understanding of what actually happens during the intervention and which PT-actions contribute to an improved developmental outcome.

Our inventory has resulted in a standardized observation protocol detailing essential elements and core features of physiotherapeutic intervention, which permitted analysis of the contents of physical therapy interventions for infants (*see Chapter 5*). The protocol was proven to meet important psychometric requirements and captured nearly all physiotherapeutic actions which were performed during intervention sessions of infants at high risk. It is an instrument which may be used to assess heterogeneity in physical therapy as it offers a tool to describe operationally what therapists do within treatment sessions across varying frames of theoretical references. Four conclusions can be drawn from the three studies (*Chapter 5-7*) which applied the observation protocol.

First, traditional infant physical therapy is characterized by a large heterogeneity in implementation. Presumably this heterogeneity is brought about by the application of techniques belonging to different treatment approaches. Not only actions belonging to the concept of Neurodevelopmental Treatment (NDT) including baby treatment - approaches well known for their heterogeneity in application (Bly 1991) - are incorporated into treatment but also actions from a more functional point of view. Research demonstrated that eclectic approaches tend to have smaller effects due to the fact that there are more confounding factors which might influence the effect size (Mandich et al 2001).

Second, the analysis of the contents of physical therapy sessions indicated a discrepancy between actual clinical practice and the physiotherapist's interpretation of his or her own actions. The evaluation of the video's taught us that physical therapists often do other things than they intend to do. This indicates that it is very important to have a uniform language within physiotherapeutic practice. Currently, different treatment approaches use the same word to describe different things. For instance, let us focus on the word "active". NDT treatment aims to give the child sensorimotor experience through the child's active responses to specialized handling (Brice, Bobath Centre, instruction video 1990). This implies that the active behaviour of the child is in part a reaction to the handling technique of the PT. In functional therapy and COPCA "active" refers to self-produced activities of the child. These frames of reference may differ more often than we realize, resulting in miscommunication. Video feedback might serve as a tool to capture this type of misunderstanding and subsequently add to the uniformity in the language used by therapists. This means that only the evaluation of real action and communication through standardized, computer-based video-analysis of treatment sessions will provide insight in the reality of practice.

Third, analysis of the newly developed programme COPCA showed many differences in the contents of COPCA and TIP (*Chapter 6*). The differences found between COPCA and TIP corresponds to the theoretical frameworks of both approaches (*see Table 6.1*). They included differences in the role of caregivers and the approach of families, the application of educational actions towards the infant and in activities to stimulate the infant's sensorimotor development. These findings indicate that COPCA and TIP based on NDT differ largely justifying the label 'novel intervention' for the COPCA programme.

Fourth, the process evaluation indicated that several aspects of the COPCA intervention were associated with an improved developmental outcome. Infants seem to benefit the most from activities that add to a challenging environment in which large variation, trial-and-error experiences and minimal postural support is offered. The physical therapist can coach the caregivers to incorporate these aspects into all daily activities. Some typical TIP actions were also associated with developmental outcome. The use of facilitation techniques like handling, which are applied to reduce atypical functional activities and to prepare the infant for optimally independent function (Dirks et al, submitted for publication), was associated with a worse performance on the functional skills scale of the PEDI. The use of sensory and passive experience showed a positive

short term effect on the cognitive development of the infants. Although the results of the VIP project seem promising, it is important to realize that the results of the study should be interpreted with caution. After all, associations do not directly mean that there is a causal relation between a PT-action and developmental outcome. Furthermore the associations between PT-actions and outcome are unavoidably contaminated with the child's initial degree of impairment. Multivariate statistics can be applied to account for this problem but still more research is needed to examine the influence of the contents of intervention on developmental outcome.

To summarize, although the differences between the COPCA and TIP group on several aspects of developmental outcome on RCT level are small, the results suggest that COPCA is more promising than TIP to influence aspects of developmental outcome. This is reflected in the findings that infants who received COPCA decline less in cognitive function over time and that COPCA possibly has a beneficial influence on postural control. The most important argument that points to a promising effect of COPCA is that key-elements of COPCA, i.e. coaching, variation, challenging, and exploration, are associated with an improved developmental outcome on the functional mobility scale of the PEDI. Hence combining the RCT with a process evaluation turned out to be the ultimate combination to examine the research questions. The possible promising effect of COPCA is in line with the results from the systematic reviews which indicated that general developmental programmes are the most promising to influence developmental outcome in infants with or at high risk for developmental motor disorders.

9.2 The VIP project; strengths and limitations

The early detection of infants at high risk for developmental motor disorders, such as cerebral palsy (CP) offers the opportunity for intervention at young age. Research showed that the best time window for early intervention in terms of active stimulation of the infant lies between 40-44 weeks post menstrual age and 15 months postnatally (De Graaf-Peters and Hadders-Algra 2006). In the VIP project the infants received intervention from three to six months which lies entirely within this recommended time window.

Selection procedure and study group

The infants were selected to participate in the VIP project when they showed definitely abnormal general movements at the age of ten weeks corrected age. Various studies indicated that the presence of definitely abnormal GM is associated with a high risk for developmental motor disorders (Prechtl 2001, Hadders-Algra 2004). It turned out that approximately 25% of the infants who had been admitted to the NICU showed definitely abnormal General Movements (GMs). The response to participate in the VIP project was 75% which resulted in a final study group of 46 infants. Chief among the reasons for families to refuse participation were the distance to the UMCG and the load and duration of the study.

One of the limitations of the VIP project was that we studied a relatively small group of 46 infants. Although power analysis before the start of the project showed that a sample size of 40 infants would be appropriate to detect differences in treatment, it is still a very small group to detect real, and clinically worthwhile, differences in treatment. This is even more complicated by the fact that is considered unethical to assign participants to a non-treatment control group (Majnemer 1998, Siebes et al 2002). Also in the VIP project we encountered this challenge. Therefore it was only possible to compare the effects of COPCA and TIP. The differences which we observed in the RCT represent only the additional value of COPCA compared to TIP. It would be interesting to know what the effects of both COPCA and TIP would be when they are compared to a group that received no intervention. These two aspects indicate that we have to be very careful with the interpretation and generalization of the results of the VIP project.

Another issue, which poses a challenge to the possibility to detect differences in developmental outcome between two groups of high risk infants, is that development in early childhood is very variable. In young infants brain dysfunction is expressed as generalized and unspecific. Infants with a left-sided intraventricular hemorrhage may respond with signs of hypotonia, hypertonia, hypokinesia or with a hyperexcitability syndrome (Hadders-Algra 2004). To make it even more complicated, brain dysfunction present at birth is not always permanent. Due to neurodevelopmental changes in the brain part of the infants with neonatal neurological symptoms may recover (Hadders-Algra 2002, 2004). Note that also the opposite is possible (Hadders-Algra 2002, 2004). This makes the developmental course of infants with a high risk for developmental motor disorders a fairly unpredictable process (Hadders-Algra 2002, Lebeer and Rijke 2003).

Evaluation and outcome measures

One of the strengths of the VIP project was that we studied the effect of intervention on short term (immediately after the intervention at 6 months CA) and on long term (one year after the intervention at 18 months CA). The outcome measures used in the VIP project to evaluate the developmental outcome were age specific and belong to the international standard. It is however acknowledged that standardized outcome measures which are often applied to measure changes in developmental outcome are in general characterized by a lack of sensitivity to detect small changes in motor or cognitive development although these small changes may have an important influence on the functional abilities of the child. The use of discriminative measures, which focus on the comparison of a child's score with an age-equivalent score, may be one of the reasons that so little effect of early intervention is found (Ketelaar et al 1998).

Neuromotor outcome was evaluated with two age specific instruments; the TINE (Touwen Infant Neurological Examination; Hadders-Algra et al 2010) from three to six months and the Hempel assessment at 18 months CA (Hempel 1993). Additionally the Neurological Optimality Score (NOS; Huisman et al 1995) was used which is known to be an excellent instrument to evaluate subtle differences in neurodevelopmental outcome (Bouwstra et al 2006). A striking finding was that the neurological outcome in the COPCA and TIP group were similar at 6 and 18 months. Considering the opposite view of COPCA and TIP on two important aspects of neurological

condition, i.e. muscle tone and abnormal movement patterns this was an interesting finding. In COPCA no attention is paid to these impairments while in TIP treatment, especially in the classic NDT variant, they play a fundamental role. It seems that therapeutic techniques aiming to influence muscle tone and movement patterns applied during early infancy do not affect neurological outcome at 18 months.

While only a minority of the infants at high risk for developmental motor disorders develops cerebral palsy (Marlow 2004), more than 50 percent shows minor neurological dysfunctions (Stahlmann et al 2007). Although neurological outcome did not differ between both groups, also the majority of the infants who participated in the VIP project (n=31) had complex minor neurological dysfunction (MND) at 18 months CA. During the intervention period most infants continued to show signs of neurological dysfunction, indicating that they were still in need of physiotherapeutic guidance. The presence of complex MND during early infancy is a risk-factor for the development of MND, DCD, attention and behavioural problems at school age (Jongmans et al 1997, Hadders-Algra 2003, 2004, Groen et al 2005).

Motor development in the VIP project was evaluated with the AIMS. One problem was that at 18 months the AIMS suffered from a ceiling effect as the majority of the infants already reached the stage of independent walking. Therefore it was not possible to differentiate between the motor outcome of the COPCA and TIP group at the follow-up. Another problem associated with the AIMS is that it mainly focuses on the observation of milestones and specific aspects of motor behaviour. What we need is a reliable, valid and sensitive instrument which in addition to motor milestones pays attention to qualitative aspects of motor development. Possibly the recently developed Infant Motor Profile (IMP; Heineman et al 2008) can solve this problem. The IMP is a video-based assessment of motor behaviour in infancy that does not only address the infant's motor abilities but also addresses variation in motor behaviour, the ability to select movement strategies, symmetry, and fluency of motor behaviour. Currently the data of the IMP for the infants who participated in the VIP project are being processed.

Cognitive outcome was assessed with the mental scale of the Bayley Scales of Infant Development (BSID). The BSID mental scale, like most developmental instruments which measure cognitive development in young children, heavily relies on fine motor skills. This will result in lower scores for motor-impaired infants. Recently an adapted Low Motor version has been developed especially for children with motor impairments in which the mental scale is less dependent of the motor capacities of the child. Ruiter (2007) showed that the children with motor impairments scored significantly higher on the Low motor version compared to the standard version of the BSID mental scale. Probably this version gives a more adequate representation of the cognitive abilities of infants with developmental motor disorders and can be used in future research.

One should keep in mind that the questionnaires we used, i.e. the KIDS, MPOC, NCSQ, carry the risk that caregivers reflect on desired rather than actual behaviour. It is furthermore important to realize what the effect might be of certain questions on the caregivers. For instance the NCSQ contains many confronting questions for parents who had experienced stressful

times when the infant was at the neonatal intensive care unit. By completing the MPOC many caregivers reported difficulties with judging the care provided by different service providers. Often they had not matching medical experiences with service providers in different settings such as the University Medical Center and a regional hospital. While participating in the VIP project they could also judge the attitude of the physiotherapist. Therefore we recommend in line with Nijhuis et al (2007) that in future research the family-centredness of different settings should be assessed separately.

Intervention

Research into the evaluation of the effectiveness of early intervention is very complex for a host of reasons. Chief among them is that physical therapy is not a specific treatment that is delivered in a standardized manner. In other words, there is no discrete dosage administered under specific, invariable procedures in conditions that are held constant. The frequency or duration could be held constant, but the procedures depend upon the therapists' skill level and specific aims and vary accordingly. While the treatment setting could be standardized, the child's family or the child's responses to the treatment could never be (Butler and Darrah 2001).

Some infants already got physical therapy before they entered the VIP project and since they - as already mentioned - continued to show signs of neurological dysfunction during the intervention period most infants also received some physical therapy sessions after the age of 6 months. After the intervention period the suggestions of the paediatrician regarding physical therapy were followed. Twenty-five of the infants (7 COPCA, 16 TIP) received more than 10 sessions after the age of 6 months, including the ten infants who developed CP. To cope with this additional heterogeneity in therapy the frequency of additional intervention sessions was taken into account in the multivariate analyses of outcome.

It turned out that TIP was characterized by a large heterogeneity in treatment application which was inherent to the evolvement of treatment techniques and theoretical assumptions over the years and to the incorporation of new treatment approaches into older ones (Bly 1991, Howle 2002, Blauw-Hospers et al 2009). This heterogeneity makes it very difficult to interpret the results because the contents of the individual treatments differ so much that the aspects which are responsible for the observed changes cannot be distinguished. The process evaluation gave us a reliable quantification of TIP in the Netherlands. It enabled us to tackle the problem of the heterogeneity and answer the question which aspects of the TIP treatment are associated with developmental outcome (*Chapter 7*).

The fact that most associations between PT-actions, in particular the typical COPCA actions, during the intervention period were associated with developmental outcome measures at 18 months CA is an interesting finding. Improvement of outcome can likely be expected when a behavioural change has occurred. In the field of early intervention this means that intervention strategies should be incorporated into daily caregiving tasks, which is one of the goals of the COPCA programme (Dirks and Hadders-Algra 2003, Dirks et al submitted).

9.3 Implications for physiotherapeutic practice and directions for future research

Paediatric physical therapy for infants at high risk for a developmental motor disorder can be considered as a field in development. No evidence is available that traditional therapy approaches, such as NeuroDevelopmental Treatment and treatment according to Vojta have a positive effect on the development of infants at high risk for developmental motor disorders (Blauw-Hospers and Hadders-Algra 2005, Blauw-Hospers et al 2007, Spittle et al 2007). This is the reason that Damiano wrote 'Based on the existing evidence the traditional approaches demand serious reconsideration by those who still advocate them' (Damiano 2006). The findings that evidence for the effectiveness of the traditional EI programmes is limited initiated important shifts in the application of therapy. The most important advances are emphasis on functionality instead of normality, the infant is an active participant of the therapy, and focus on family dynamics. Currently, evidence is accumulating that intervention from a more functional approach focusing on general development is most promising to influence developmental outcome in a beneficial way (*Chapter 3 and 4*). Research has indicated that the involvement of caregivers can be associated with positive effects both on the development of the child (Ketelaar et al 1998, Law et al 1998, King et al 2004) as well as on the caregivers in terms of an increase in parental skills, improved well-being and a decreased level of parental stress (King et al 1996, Rosenbaum et al 1998, Jansen et al 2003, King et al 2004, Raina et al 2005). Another factor which may add to the possibility that intervention will sort a long term effect is the implementation of intervention strategies into daily activities. In the COPCA intervention focus is on family function, family autonomy and activities and participation in daily life activities. The results of the VIP project suggest that this strategy may be effective.

In this thesis the first results on the implementation of the COPCA programme are described. Although the effects on RCT level were small, the results from the process evaluation showed that key aspects of COPCA, i.e. family involvement and educational actions, application of large variation in challenging the infant to produce motor behavior by him/herself and allow the infant to continue this activity, and stimulation of motor behavior at the verge of the infant's capabilities, were associated with improvements in developmental outcome at 18 months (*Chapter 7*).
To establish real insight in the effects and working mechanisms of COPCA on developmental outcome more research has to be performed. It is likely that the effect of intervention is a combination of several factors which are related to the condition of the infant, the family, the therapist, the contents of the intervention, and environmental factors. As some problems related to early neurological dysfunction become more visible at school age future studies should include a longer follow-up.

We described a first step in objectifying physiotherapeutic practice. By uncovering the contents of infant physical therapy in the Netherlands we obtained insight into which actions constitute practice and we were able to study relations with developmental outcome. A next step in objectifying physiotherapeutic practice might be a study on the similarities and differences in the application of infant physical therapy in other countries. Additionally more research must be performed to optimize the psychometric requirements of the observation protocol.

Future research should not only focus on the development of new intervention strategies. Adequate assessment techniques are also indispensable for the evaluation of the effectiveness of intervention. There is a need for more scientific sound qualitative instruments which are able to assess and evaluate the developmental outcome of infants at high risk for developmental motor disorders more accurately. Current advances show that the psychometric properties of several qualitative instruments are being examined (for instance the Infant Motor Profile (Heineman et al 2008), the Touwen Infant Neurological Examination (Hadders-Algra et al 2010), and the Low Motor Version of the Bayley Scales of Infant Development (Ruiter 2007)) suggesting that these methods can be applied in future studies.

The implementation and evaluation of a new outcome measure or intervention programme into clinical practice takes several years (Ketelaar et al 2008). Here a possible threat comes up. As physiotherapists tend to include only techniques they perceive as helpful into their treatment, the risk exists that a new intervention is not implemented in the way it was intended. Good education is essential to diminish this threat. Although the results are still premature and more research has to be performed, COPCA - a family friendly approach to intervention - has the potential to influence developmental outcome of infants with high risk for developmental motor disorders in a beneficial way. Currently another trial on the effectiveness of COPCA is carried out: Learn to Move 0-2 years (L2M0-2). L2M0-2 is part of a multicenter trial on the effectiveness of intervention in children and adolescents with CP. In L2M0-2 many of the limitations encountered in the VIP project are accounted for. In the near future this will result in more knowledge about the effect of early intervention on the developmental outcome of infants at high risk for developmental motor disorders.

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SUMMARY

Young infants, who are at risk for developmental motor disorders, such as cerebral palsy (CP) or developmental coordination disorder, are often referred to early intervention services. The field of early intervention consists of multidisciplinary developmental, educational and therapeutic services provided to children from birth to 5 years of age. In this thesis early intervention is restricted to paediatric physical therapy for infants up to 18 months corrected age (CA). The introduction (*Chapter* 1) starts with an overview of the main influences on paediatric physical therapy in the Netherlands. Paediatric physical therapy for infants at high risk for a developmental motor disorder can be considered as a field in development. Based on findings that evidence for the effectiveness of the traditional early intervention programmes (NeuroDevelopmental Treatment, treatment according to Vojta, and Sensory Integration Therapy) is limited, shifts in the application of therapy were initiated. The most important advances are a focus on functionality instead of normality, the infant as an active participant of the therapy and the inclusion of the family.

The first aim of this thesis was to review the effects of early physiotherapeutic intervention on motor and cognitive development of infants with a high risk for developmental motor disorders aged less than 18 months (*Chapter 3 and 4*). The reviews showed that the effect of early intervention programmes which focus primarily on the improvement of motor development is inconclusive. It turned out that the type of intervention which might be beneficial for infants at preterm age differs from the type which is effective for infants who have reached at least term age. At preterm age infants seem to benefit most from intervention which aims at mimicking the intrauterine environment. After term age, general developmental programmes in which parents learn how to promote infant development can exert a positive effect on motor and cognitive development. Traditional therapy approaches, such as NeuroDevelopmental Treatment and treatment according to Vojta, do not influence the developmental outcome of infants at high risk for developmental motor disorders.

The results of both reviews indicated that intervention programmes with an onset after term age which focus on the stimulation of self-produced motor behaviour may exert a beneficial effect on motor and cognitive development. This knowledge served as one of bases for the development of a new type of physiotherapeutic intervention, COPCA. COPCA is the abbreviation of Coping with and Caring for infants with special needs – a family centred programme. It is a family relationship oriented programme consisting of two theoretical constructs. The first component is formed by a family involvement and educational parenting component. The second theoretical component, a neurodevelopmental one, is based on the principles of the Neuronal Group Selection Theory.

The second aim of this thesis was to evaluate the effects of the COPCA intervention on developmental outcome. In the early intervention project VIP (in Dutch "vroegtijdig interventie project";

Chapter 2) 46 high-risk infants were randomly assigned to receive COPCA (n=21) or traditional infant physical therapy (TIP; n=25) between 3 to 6 months CA. Developmental status of the infants was assessed at 3, 4, 5, 6 and 18 months CA with instruments measuring at impairment level (neurological examination) to instruments on the level of activity and participation (PEDI).

The randomized controlled trial (RCT) revealed that developmental outcome in both groups was largely identical (*Chapter 7*), except for cognitive outcome. Ten infants were diagnosed with CP at the age of 18 months CA. The majority of infants (15 COPCA, 16 TIP) showed complex minor neurological dysfunction, which puts the infant at risk for learning and behavioural disorders at school age. The RCT had two interesting results. First, there was an interaction effect between the type of intervention and the level of maternal education in the relative drop in MDI score between 6 and 18 months CA. Infants whose mother had a lower level of education declined less in MDI score over time compared to infants whose mother had finished a higher education. This effect was in particular present in the COPCA group. This suggests that in particular mothers with a lower education benefit more from coaching by a therapist who uses COPCA principles, while mothers with a higher education are less affected by coaching as well as training performed by a therapist. Second, the finding of a similar neurological outcome in the COPCA and TIP group is intriguing considering the opposed view of TIP and COPCA on the importance of the neurological parameters muscle tone and atypical movements. In the TIP treatment, especially when applied to the original concepts of Bobath and/or NDT, tone influencing by means of handling techniques plays an important role in the activities of the therapist during intervention sessions. In COPCA no attention is paid to these impairments. In other words, handling techniques aiming to influence muscle tone and facilitating movement sequences to improve function, do not seem to affect neurological outcome.

Some supplementary findings of the VIP project are discussed in Chapter 8. Other data of the VIP project are currently analysed (motor development in terms of the Infant Motor Profile and postural control, and video-recordings of daily life activities).

The third aim of this thesis was to evaluate the application of COPCA in daily practice by means of video analysis. This process evaluation is a prerequisite for the implementation of COPCA within paediatric physiotherapy. The first step to achieve this was the development of a standardized, theory based, observation protocol detailing essential elements and core features of paediatric physiotherapeutic intervention, which permitted analysis of the content of physical therapy interventions for infants (*Chapter 5*). The observation protocol classifies PT actions into eight mutually exclusive categories: A) Family involvement and educational actions, B) Communication , C) Facilitation techniques, D) Sensory experience, E) Passive motor experience, F) Self-produced motor behaviour, no interference with PT /caregiver, G) Challenge to self-produce motor behaviour by him/herself; infant is allowed to continue activity, H) Challenge to self-produce motor behaviour by him/herself; activity flows over into therapeutic handling. The protocol was proven to meet important psychometric requirements and captured nearly all physiotherapeutic actions

which were performed during intervention sessions of infants at high risk for a developmental motor disorder. The observation protocol may be used to assess heterogeneity in physical therapy as it offers a tool to describe operationally what therapists do within treatment sessions across varying frames of theoretical references. Four conclusions can be drawn from the three studies (*Chapter 5-7*) which applied the observation protocol:

1) Traditional infant physical therapy is characterized by a large heterogeneity in implementation which probably is caused by the application of techniques belonging to different treatment approaches.
2) A discrepancy exists between actual clinical practice and the physiotherapist's interpretation of his or her own actions.
3) The contents of COPCA and TIP sessions differed largely, suggesting that it is possible to implement COPCA in daily practice by physical therapists who received special training in the COPCA programme (*Chapter 6*). The differences found between COPCA and TIP corresponds to the theoretical frameworks of both approaches. They included differences in the role of caregivers and the approach of families, in the application of educational actions towards the infant and in activities to stimulate the infant's sensorimotor development.
4) Several aspects of the COPCA intervention, i.e. family involvement and educational actions, application of large variation in challenging the infant to produce motor behaviour by him/herself and allow the infant to continue this activity, and stimulation of motor behaviour at the verge of the infant's capabilities, were associated with an improved developmental outcome. The use of facilitation techniques like handling was associated with a worse performance on the functional skills scale of the PEDI. The use of sensory and passive experience was associated with a short term positive effect on cognitive development.

It is important to realize that the results of this study should be interpreted with caution as the observed associations do not automatically mean that a causal relation between contents and outcome exists. Associations between PT actions and outcome are unavoidably contaminated with the child's initial degree of impairment. Multivariate statistics were applied to account for this problem but still more research is needed to examine the influence of the contents of intervention on developmental outcome.

In *Chapter 9* the main findings of this thesis are summarised and discussed and placed into perspective. Although the differences between the COPCA and TIP group on several aspects of developmental outcome on RCT level are small, the results suggest that COPCA - a family friendly approach to intervention - has the potential to influence developmental outcome of infants with high risk for developmental motor disorders in a beneficial way. The most important argument that points to a promising effect of COPCA is that key elements of COPCA, i.e. coaching, variation, challenging, and exploration, are associated with an improved developmental outcome. Hence combining the RCT with a process evaluation turned out to be the ultimate combination to examine the research questions. The possible promising effect of COPCA is in line with the

results from the systematic reviews which indicated that general developmental programmes are the most promising to influence developmental outcome in infants with or at high risk for developmental motor disorders.

In the next years the multicenter trial Learn to Move 0-2 years will provide more knowledge about the effect of early intervention on the developmental outcome of infants at high risk for cerebral palsy in general and the COPCA programme in particular.

SAMENVATTING

Jonge kinderen met een verhoogd risico op het krijgen van een motorische ontwikkelingsstoornis, zoals cerebrale parese (CP) of developmental coordination disorder, komen in aanmerking voor vroegtijdige interventie. De term vroegtijdige interventie omvat een scala aan multidisciplinaire therapeutische mogelijkheden die zich richten op de ontwikkeling en opvoeding van kinderen tussen nul en vijf jaar. In Nederland worden kinderen met een verhoogd risico op het krijgen van een motorische ontwikkelingsstoornis vaak verwezen naar de kinderfysiotherapeut. In dit proefschrift is vroegtijdige interventie afgebakend tot kinderfysiotherapie voor kinderen jonger dan de gecorrigeerde leeftijd van 18 maanden. De introductie (*Hoofdstuk 1*) geeft een overzicht van de belangrijkste ontwikkelingen binnen de kinderfysiotherapie in Nederland. Kinderfysiotherapie is een discipline die zich de afgelopen decennia sterk heeft ontwikkeld. Er zijn een aantal belangrijke verschuivingen opgetreden. De nadruk ligt tegenwoordig op functionaliteit in plaats van normale bewegingspatronen. Ook wordt het gezin actief betrokken bij het therapeutisch proces en neemt het kind actief deel aan zijn of haar eigen therapie.

Het eerste doel van dit proefschrift was het in kaart brengen van de bestaande effecten van vroegtijdige fysiotherapeutische interventie op de motorische en cognitieve ontwikkeling van kinderen die jonger zijn dan 18 maanden en een verhoogd risico hebben op het krijgen van een motorische ontwikkelingsstoornis (*Hoofdstuk 3 en 4*). Uit de beide systematische literatuurstudies blijkt dat het effect van interventies die zich primair richten op het verbeteren van de motorische ontwikkeling onduidelijk is. Het type interventie dat mogelijk effect heeft, verschilt voor kinderen op de preterme leeftijd en kinderen die tenminste de à terme leeftijd bereikt hebben. Voor de à terme leeftijd lijken kinderen het meest baat te hebben bij interventies waarin de situatie van de baarmoeder wordt nagebootst. Na de à terme leeftijd hebben programma's die zich richten op de algemene ontwikkeling van het kind en waarin ouders leren hoe ze de ontwikkeling van hun kind kunnen stimuleren de meeste potentie. De traditionele benaderingen, zoals NeuroDevelopmental Treatment en behandeling volgens de methode Vojta, hebben geen effect op de ontwikkeling van kinderen met een verhoogd risico op het krijgen van een motorische ontwikkelingsstoornis.

De resultaten van beide literatuurstudies laten zien dat interventie programma's, die starten na de à terme leeftijd en zich richten op het stimuleren van zelf geproduceerd motorisch gedrag, de motorische en cognitieve ontwikkeling positief kunnen beïnvloeden. Deze kennis heeft bijgedragen aan de ontwikkeling van een nieuwe vorm van kinderfysiotherapeutische interventie genaamd COPCA (Coping with and Caring for infants with special needs - a family-centered programme). COPCA is gericht op de relaties en interacties binnen het gezin. Het programma is gebaseerd op nieuwe inzichten op het gebied van pedagogiek en gezinsgerichte zorg en op de principes van de Neuronale Groep Selectie Theorie.

Het tweede doel van dit proefschrift was het evalueren van het effect van COPCA op de ontwikkeling van kinderen met een verhoogd risico op het krijgen van een motorische ontwikkelingsstoornis. Aan het Vroegtijdig Interventie Project (VIP; *Hoofdstuk 2*) hebben 46 kinderen deelgenomen. De kinderen zijn via blokrandomisatie verdeeld over de experimentele COPCA groep (21 kinderen) en de controlegroep (25 kinderen) die de traditionele vorm van kinderfysiotherapeutische begeleiding (TIP) op indicatie van de kinderarts kreeg. De interventieperiode duurde van 3 tot 6 maanden. De ontwikkeling van de kinderen is gemeten op de gecorrigeerde leeftijd van 3, 4, 5, 6 en 18 maanden aan de hand van meetinstrumenten die zich richten op de neuromotore ontwikkeling, het psychosociaal functioneren en de neurofysiologische parameters van de houdingsregulatie.

Uit de analyses kwam naar voren dat er op groepsniveau (RCT) weinig verschillen waren tussen de COPCA en de TIP groep (*Hoofdstuk 7*), behalve in de cognitieve ontwikkeling. Tien kinderen hadden een CP ontwikkeld op de gecorrigeerde leeftijd van 18 maanden. Het merendeel van de kinderen (15 COPCA, 16 TIP) liet de complexe vorm van minor neurological dysfunction zien, waardoor ze op de schoolleeftijd meer risico lopen op leer- en gedragsproblemen. Twee interessante resultaten kwamen uit het RCT naar boven. Ten eerste bestond er een interactie tussen het opleidingsniveau van de moeder en het type interventie met de verandering in de cognitieve uitkomst tussen 6 en 18 maanden. Dit betekende dat kinderen van moeders met een lagere opleiding uit de COPCA groep minder achteruitgingen in hun MDI score dan andere kinderen. Dit suggereert dat moeders met een lager opleidingsniveau meer profiteren van het coachen door een therapeut die de COPCA methode gebruikt. Ten tweede viel op dat de neurologische uitkomst in de COPCA en de TIP groep vergelijkbaar was. Dit is interessant gezien de tegenovergestelde kijk van TIP en COPCA op neurologische parameters zoals spierspanning en afwijkende bewegingen. In TIP vormen tonus beïnvloedende strategieën een belangrijke rol in de fysiotherapeutische acties, terwijl COPCA hier geen aandacht aan besteed. Oftewel, fysiotherapeutische technieken gericht op het beïnvloeden van de spierspanning en het faciliteren van 'normale' bewegingspatronen lijken geen effect te hebben op de neurologische ontwikkeling. In *Hoofdstuk 8* staan een aantal aanvullende resultaten van meetinstrumenten uit het VIP project beschreven die niet gepubliceerd zijn. Andere uitkomsten worden momenteel geanalyseerd, zoals de motorische ontwikkeling in termen van de Infant Motor Profile, parameters van de houdingsregulatie en video opnames van activiteiten in het dagelijks leven (baden en spelen).

Het derde doel van dit proefschrift was de evaluatie van de toepasbaarheid van COPCA in de dagelijkse praktijk. Daarvoor zijn video opnames gemaakt van fysiotherapeutische behandelingen op 4 en 6 maanden. Om de inhoud van de fysiotherapeutische behandeling te kunnen analyseren, is eerst een gestandaardiseerd observatie protocol ontwikkeld (*Hoofdstuk 5*). Dit protocol is opgebouwd uit de belangrijkste elementen en kern items van kinderfysiotherapeutische interventie. Fysiotherapeutische handelingen worden ingedeeld in 8 categorieën: A) Mate van betrokkenheid van het gezin en opvoedingselementen gericht op het kind/gezin, B) Communicatie, C) Facilitatie technieken, D) Sensorische stimulatie, E) Passief bewegen, F) Zelf geproduceerd motorisch gedrag zonder tussenkomst van de fysiotherapeut of ouder G) Uitgelokt zelf geproduceerd motorisch gedrag dat wordt voortgezet door het kind, H) Uitgelokt zelf geproduceerd motorisch gedrag dat overgaat in handling. Analyses lieten zien dat het protocol voldoet aan belangrijke psychometrische eisen zoals een voldoende inter- en intrabeoordelaarsbetrouwbaarheid en dat bijna alle fysiotherapeutische handelingen gescoord kunnen worden in de categorieën van het protocol. Het observatieprotocol kan gebruikt worden als instrument om de heterogeniteit in de fysiotherapie te meten. Het geeft een gedetailleerd overzicht van de handelingen die een kinderfysiotherapeut toepast binnen zijn of haar behandeling en houdt tegelijkertijd rekening met verschillende theoretische referentiekaders. Uit de studies waarin het observatieprotocol toegepast is (*Hoofdstuk 5-7*), kunnen vier conclusies worden getrokken:

1) Traditionele kinderfysiotherapeutische begeleiding wordt gekenmerkt door een zeer heterogene toepassing. Dit wordt mogelijk veroorzaakt door het integreren van technieken uit verschillende stromingen binnen één behandeling.
2) Er bestaat een discrepantie tussen de werkelijke fysiotherapeutische handelingen en de interpretatie van de kinderfysiotherapeut over de handelingen die hij/zij toepast.
3) De inhoud van COPCA en TIP is zeer verschillend (*Hoofdstuk 6*). Dit suggereert dat het mogelijk is om COPCA te implementeren in de dagelijkse praktijk, mits het gegeven wordt door fysiotherapeuten die een formele training in de COPCA methode gevolgd hebben. De verschillen tussen COPCA en TIP corresponderen met de theoretische kaders van beide interventieprogramma's. Er bestaan verschillen in de rol van de ouders en de benadering van de familie, in de opvoedingselementen gericht op het kind en in activiteiten die zich richten op het stimuleren van de sensomotorische ontwikkeling van het kind.
4) De inhoud van de behandeling speelt wel degelijk een rol bij de ontwikkeling van kinderen met een hoog risico op een motorische ontwikkelingsstoornis. Een aantal aspecten van de COPCA interventie , namelijk de mate van betrokkenheid van de familie en opvoedingselementen gericht op het kind/gezin (met name coaching), het uitlokken van zelf geproduceerd motorisch gedrag dat wordt voortgezet door het kind met veel variatie en het stimuleren van motorisch gedrag op de grens van de mogelijkheden die een kind heeft, zijn geassocieerd met een betere ontwikkelingsuitkomst. Het gebruik van facilitatie technieken, zoals handling, was geassocieerd met een slechtere uitkomst op de functionele vaardigheden schaal van de PEDI. Sensorische stimulatie en passief bewegen waren op de korte termijn geassocieerd met een betere cognitieve ontwikkeling, maar dit verband was niet meer aanwezig op 18 maanden. Het is belangrijk om te beseffen dat de resultaten van deze studie voorzichtig geïnterpreteerd dienen te worden aangezien het bestaan van een associatie niet automatisch betekent dat er een causale relatie bestaat tussen de inhoud en de uitkomstmaat. Associaties tussen fysiotherapeutische handelingen en ontwikkeling zijn onvermijdelijk gecontamineerd met de uitgangssituatie van het kind. Om dit probleem te ondervangen hebben we gebruik gemaakt van multivariate statistiek, maar er is meer onderzoek nodig om de invloed van de inhoud van fysiotherapeutische interventie op de ontwikkeling vast te kunnen stellen.

In *Hoofdstuk 9* wordt een overzicht gegeven van de belangrijkste bevindingen uit dit proefschrift. Alhoewel de verschillen tussen de COPCA en de TIP groep op groepsniveau klein zijn, lijkt het erop dat COPCA mogelijk de ontwikkeling van kinderen die een verhoogd risico hebben op het ontwikkelen van een motorische ontwikkelings-stoornis positief kan beïnvloeden. Deze gedachte wordt m.n. ondersteund door de bevinding dat een aantal kernitems van COPCA, namelijk coachen, variëren, uitdagen en exploreren, geassocieerd zijn met een betere ontwikkeling. We concludeerden dan ook dat het uitbreiden van de RCT met een procesevaluatie de mogelijkheid schepte om de onderzoeksvragen goed te kunnen beantwoorden. Het mogelijke positieve effect van COPCA komt overeen met de resultaten van de systematische literatuurstudies waaruit blijkt dat vroegtijdige interventie programma's, die zich richten op de algehele ontwikkeling van het kind en waarin ouders leren hoe ze de ontwikkeling van hun kind kunnen stimuleren, de ontwikkeling van kinderen met een verhoogd risico op het krijgen van een motorische ontwikkelingsstoornis positief kunnen beïnvloeden.

In de komende jaren zal het onderzoek Learn to Move 0-2 jaar meer inzicht verschaffen in het effect van vroegtijdige interventie op de ontwikkeling van kinderen met een hoog risico op cerebrale parese in het algemeen en in het COPCA programma in het bijzonder.

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ABOUT THE AUTHOR

Cornill Blauw-Hospers werd geboren op 8 januari 1978 te Groningen. Na het behalen van haar VWO diploma aan het Ubbo Emmius Lyceum te Stadskanaal in 1996 startte zij met de opleiding Bewegingswetenschappen aan de Rijksuniversiteit Groningen. In 2000 studeerde zij af met als specialisatie Sport en Lichamelijke opvoeding. Haar afstudeerproject betrof de evaluatie van het effect van fysiotherapie voor kinderen met motorische problemen. Na haar afstuderen werkte zij enige tijd als onderzoeker bij het Centrum voor Revalidatie Beatrixoord van het Universitair Medisch Centrum Groningen. Na een verhuizing naar Sneek zette zij haar onderzoeksloopbaan voort in Groningen waar zij op 1 maart 2003 werd aangesteld als AIO bij het Instituut voor Ontwikkelingsneurologie van het UMCG. Zij deed daar onderzoek naar de effecten van vroegtijdige fysiotherapeutische interventie voor jonge kinderen met een hoog risico op een motorische ontwikkelingsstoornis. De resultaten van haar studie staan beschreven in deze dissertatie. Vanaf september 2008 is zij werkzaam bij het Koninklijk Nederlands Genootschap Fysiotherapie te Heerenveen.

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APPENDIX

Observation protocol: classification of pediatric physical therapy (PT) actions

The PT actions are classified into 8 main categories which contain various subcategories (second level of observation). For each specific action one or more examples of concrete PT or caregiver actions are provided (third level of observation)

A. Family involvement and educational actions
The extent to which the family is involved in the treatment of the infant and the extent of guidance, interference, coaching or training by the physical therapist or the caregiver during the treatment session.

A.1 Family members participating in guidance infant*:
Family members involved in the treatment session.
Examples of concrete actions:
- Mother present only
- Father present only
- Both caregiver but no other family members present
- In addition to parent(s) also other family members present

A.2 Role of caregiver*:
The extent to which the family is involved in the treatment.

Examples of concrete actions:
- PT performs treatment by means of handling techniques
- Caregiver and PT act together in handling techniques, PT performs the treatment (hands on) while the caregiver guides the attention of the infant.
- Caregiver performs handling techniques thereby controlling the infant's actions. The PT instructs the caregiver how to handle
- Caregiver and PT act together (hands off), caregiver is playing with the child and may provide the infant with minimal support but leaves the infant always with ample opportunities for exploration. PT observes the caregiver-infant relationship and may give hints.
- Caregiver is playing with the infant (hands off) and leaves the infant with ample opportunities for exploration

A.3 Infant dressing*:
The way in which the infant is dressed during the treatment session.
Examples of dressing:
- Infant is dressed
- Infant is partially dressed
- Infant is undressed (wearing underwear only)

A.4 Educational actions
A.4.1 The extent of interference by the PT or the caregiver with infant's activities
Examples of concrete actions:
- PT or caregiver interrupts activities of the infant.
- PT or caregiver corrects when infant fails.
- PT or caregiver assists when infant has difficulties to perform an action

A.4.2 The extent of guidance the infant by the PT.
Examples of concrete actions:
- PT trains motor performance at the level which is relatively too easy for the infant.
- PT provides excessive postural support or assistance.
- PT trains motor performances at the level which is too difficult for the infant.

A.4.3. Caregiver training
All actions during which the PT instructs caregivers how to handle the infant with the aim that caregivers can continue treatment strategies during daily life activities and/or in the home environment.
Examples of concrete actions:
- PT demonstrates therapeutic handling action to caregiver.
- PT demonstrates action to caregiver, variable options provided.
- PT practices with caregivers teaching them how to continue some of handling techniques in daily life at home.

A.4.4 Caregiver coaching
All actions during which the PT coaches the caregiver. Coaching aims to empower caregivers so that they can made their own educational decisions during daily care activities in the home environment.

Examples of concrete actions:
- Caregiver patiently observes the infant actions
- Caregiver challenges motor performances just at the verge of infant's abilities.
- Caregiver provides as little postural support as possible – challenges postural behavior of the infant
- Caregiver tries to evoke pleasure in the infant

B. Communication
All communication between the PT and the caregiver that is related to the guidance of infant and family

B.1 Information exchange:
All communication during which information is exchanged regarding experiences, worries and the role of the PT.
Examples of concrete actions:
- PT provides opportunity for caregivers to tell about experiences related to NICU stay, to express worries and concerns about infant and family matters.
- PT provides information about role of PT and caregiver.
- PT asks whether specific problems exist.

B.2 Contents of information:
All communication that explains the background of the treatment strategies, including developmental education.
Examples of concrete actions:
- PT explains handling in terms of typical movement patterns, typical development, posture, muscle tone, asymmetry/symmetry and hand placing.
- PT explains the need of, variation, minimal support, exploration, trial and error, challenge, patience.
- PT [explains the need to explore communication.
- PT discusses the application of intervention strategies into daily routines in terms of handling.
- PT discusses the application of the intervention into daily routines in terms of variation, exploration, motor challenge

B.3 Instruct:
All communication in which the caregiver is given assignments or hints regarding treatment strategies.
Examples of concrete actions:
- PT assigns, gives advice what to do.
- PT gives hints, provides a suggestion or clue in a very indirect way so that caregivers feel free to explore ample variable opportunities.

B.4 Provide Feedback:
All communication in which the treatment or the performances of infant and caregiver are evaluated.
Examples of concrete actions:
- PT tells the caregiver what went right/wrong.
- PT evaluates the procedure.
- PT asks and listens to the opinion of the caregiver.

B.5 Impart knowledge:
All communication that provides the caregiver with knowledge about the therapeutic actions that are performed.
Examples of concrete actions:
- PT asks about performance action.
- PT explains the ins and outs of an action.
- PT asks about understanding.
- PT asks about ability of caregiver to perform an action and listen to caregivers comments on actions.

C. Facilitation techniques.
All therapeutic hands on actions of the physical therapist or caregiver aiming at guidance of the movement or maintenance of the infant's posture by gently placing the hands on specific parts of the infant's body, thus providing the infant with sensorimotor experience and controlling movement output.

C1. Handling:
Specific hands-on techniques to give the infant sensorimotor experience to improve the quality and repertoire of the infant's movements.
Examples of concrete PT/caregiver actions:
- In supine position or in sitting. Shoulders function as key point: handling hands guide shoulders of the infant in protraction to control tone and to facilitate hand-hand contact and symmetry.
- In supine position. Proximal or distal leg functions as key point: the infant's hip is passively brought in semi-flexion while adducting the leg across the midline to facilitate head righting and rolling.

- In supine position. Pelvis functions as key point: the infant's pelvis is slightly lifted to elongate the extensor muscles of the trunk and to control tone; in this way hand-foot contact and symmetry are facilitated
- In prone position. Shoulder functions as key point, the arms are placed in puppy position to facilitate head righting, midline orientation and body-alignment.
- In sitting. Shoulder functions as key point: the shoulders are moved alternately forwards and backwards to dissociate and facilitate independent arm movements.

C.2 Pressure techniques:
All handling techniques that produce intermittent pressure to stimulate and gain control over muscle tone, posture and movement.
Examples of concrete PT /caregiver actions:
- In sitting: intermittent downward pressure on shoulders in the direction of the pelvis to facilitate extension of the trunk
- In sitting: slight intermittent pressure movements on abdominal region in direction of the sacrum to facilitate contraction of the ventral muscles.

C.3 Transition:
All handling techniques that result in the change of position of the infant.
Examples of concrete PT /caregiver actions:
- From supine to side, from supine to prone, from supine to sit, from side to sit, from prone to supine, from sit to supine, etc.

C.4 Support devices:
All handling techniques which use additional devices to support the infant.
Examples of concrete / additional devices:
- Bolster or ball
- Supporting sling.

D. Sensory experience
All tactile and vestibular stimulation given to the infant during treatment - without the aim of facilitation, tapping or passive motor experience - offered to the infant to provide him/her with the perception of body awareness.
Examples of concrete PT /caregiver actions:
- Touching skin with toy.
- Tickling.
- Tapping on muscles

E. Passive motor experience
All handling techniques induced by the PT or the caregiver in which no activity of the infant is required in the performance of the actions.
Examples of concrete PT /caregiver actions:
- Passive movements of arms.
- Repetitive movements of the upper arm towards (frontal) support surface.
- Passive rocking, small sideways movements.

F. Self-produced motor behavior, no interference with PT /caregiver
All actions during which the infant is given ample opportunities to explore toys or other aspects of the environment or his body without interference of PT or caregiver.
Examples of concrete PT /caregiver actions:
- Placing an infant activity play center over the infant and let the infant explore the effect of movements or arms, hands, legs and feet.
- Infant is given opportunity for spontaneous exploration with or without toy.
- Postural challenges, infant spontaneously explores postural capacities.

G. Infant is challenged to produce motor behavior by him/herself; infant is allowed to continue activity by her/himself
All actions in which the infant is challenged by toys or face of PT or caregiver to experience a variety of motor activity, that is continued by the infant her/himself.

G.1 Little variation:
All actions in which the infant is challenged by toys or face of PT or caregiver to explore one strategy to reach and grasp, to control posture, to roll, etc.

G.2 Large variation:
All actions in which the infant is challenged by toys or face of PT or caregiver the infant is challenged to explore multiple strategies to reach and grasp, to control posture, to roll, etc.

H. Infant is challenged to produce motor behavior by him/herself; activity flows over into therapeutic handling
All actions in which the infant is challenged by toys or face of PT or caregiver to experience a variety of motor activity, that is followed by a handling technique.

H.1 Little variation:
All actions in which the infant is challenged by toys or face of physical therapist or caregiver to explore one strategy to reach and grasp, to control posture, to roll, etc.

H.2 Large variation:
All actions in which the infant is challenged by toys or face of physical therapist or caregiver the infant is challenged to explore multiple strategies to reach and grasp, to control posture, to roll, etc.

I. Not specified
All time during the treatment session that can not be classified into the eight defined categories.
Examples:
- Comforting the infant.
- Change the treatment situation.

Postural support in prone, side and sitting position:
No postural support:
PT or caregiver leaves it to the infant to adjust posture independently. "Hands off"
Minimal postural support:
PT or caregiver provides as little support as possible in order to challenge postural behavior of the infant
Example of concrete action:
PT or caregiver challenges motor performance just at the verge of the infant's abilities, i.e. the infant has to 'work' to maintain balance
Clear postural support:
PT or caregiver provides support on multiple parts of body or the trunk. Minimal active involvement of the infant to adjust posture is required.
Example of concrete action:
- PT or caregiver provides support at the neck/shoulder girdle and/or upper part of the trunk

Full postural support:
PT or caregiver supports all parts of the body of the infant that play a role in postural adjustments. No active involvement of the infant is required.

***** Independent variable: the value of this variable is supposed not to change during the course of an observation. It gives the observer the opportunity to summarize briefly important characteristics of the observation