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## Effect of early intervention on functional outcome at school age: Follow-up and process evaluation of a randomised controlled trial in infants at risk



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### ABSTRACT

**Background:** The long-term effect of early intervention in infants at risk for developmental disorders is unclear. The VIP project ( $n = 46$ , originally) evaluated by means of a randomised controlled trial the effect of the family centred early intervention programme COPCA (Coping with and Caring for infants with special needs) in comparison to that of traditional infant physiotherapy (TIP).

**Aims:** To evaluate the effect of early intervention on functional outcome at school age.

**Methods and procedures:** Parents of 40 children (median age 8.3 years) participated in this follow-up study. Outcome was assessed with a standardised parental interview (Vineland Adaptive Behaviour Scale) and questionnaires (Developmental Coordination Disorder Questionnaire, Child Behaviour Checklist, Utrechtse Coping List, and questions on educational approach). Quantified video information on physiotherapeutic actions during infancy was available.

**Outcomes and results:** Child functional outcome in the two randomised groups was similar. Process evaluation revealed that some physiotherapeutic actions were associated with child mobility and parental educational approach at follow-up: e.g., training and instructing were associated with worse mobility.

**Conclusions and implications:** Functional outcome at school age after early intervention with COPCA is similar to that after TIP. However, some specific physiotherapeutic actions, in particular the physiotherapist's approach, are associated with outcome.

**What this paper adds:** Early intervention is generally applied in infants at risk for developing disorders, with the aim of improving overall functional outcome. However, little is known on the long-term effect.

The VIP project evaluated by means of a randomised controlled trial the effect of the family centred early intervention programme COPCA (Coping with and Caring for infants with special needs) in comparison to that of traditional infant physical therapy (TIP). Outcome at 18 months corrected age was virtually similar. Process evaluation of the physiotherapy actions revealed that some characteristics of COPCA were associated with improved developmental outcome at 18 months.

This paper presents data on functional outcome at school age (median 8.3 years) in 87% of the original participants. Outcome of infants who received three months of COPCA and that of infants who received three months of TIP was similar. Yet, parents of families who had received the COPCA intervention still more often used a trial and error approach when the child learned a new skill than parents of children who had received TIP. Process evaluation showed that more time spent on caregiver training and strict instructions during early intervention was associated with worse mobility. Four other physiotherapeutic actions were associated with parental educational approach. None of the neuromotor actions were associated with child outcome at school age.

We conclude that long-term outcome after three months of COPCA or TIP is similar. However, our study does suggest that the professional approach of the physiotherapist can make a difference.

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## 1. Introduction

The long-term effect of early intervention in infants at risk for developmental disorders remains unclear. A recent Cochrane meta-analysis on early intervention in preterm born children demonstrated a small positive effect of early intervention on motor and cognitive outcome in infancy, with the cognitive effect persisting into preschool age [1]. Only a few studies evaluated developmental outcome after pre-school age [1]. The data available suggest no or inconclusive effects of early intervention. The present study aims to contribute to the limited knowledge on the effect of early intervention on developmental outcome at school age.

One of the factors that might explain the small effect of early intervention on developmental outcome is our limited understanding of which elements of intervention are effective in promoting better outcome [1–3]. It has been assumed that general developmental programmes and parental coaching are most effective [2,4]. In line with these suggestions of the literature, the family centred COPCA (COPing with and Caring for infants with special needs) programme had been developed [5,6]. Strengthening of family autonomy and participation, and promotion of infant mobility are the major goals of the COPCA programme. COPCA focuses on the family and includes educational components. The neurodevelopmental component of COPCA is based on the neuronal group selection theory (NGST) [7,8].

The VIP project (Dutch: Vroegtijdig Interventie Project) evaluated, by means of a two arm randomised controlled trial (RCT), the effect of 3 months of COPCA in early infancy in comparison to that of 3 months of traditional infant physiotherapy (TIP) in infants at risk for developmental disorders. The at risk infants had been admitted to the neonatal intensive care unit (NICU) of the University Medical Centre Groningen and showed definitely abnormal general movements at 10 weeks corrected age (CA), indicating that they had a high risk for developmental disorders like cerebral palsy (CP). As paediatric physiotherapy is characterized by heterogeneity, we presumed that the contents of the two intervention programmes would overlap. We therefore had expected that the difference in outcome of the two randomised groups might be minimal. Indeed, at 18 months CA we only found a minor advantage of COPCA for cognitive development, when the level of maternal education was taken into account [9]. In anticipation, we therefore had video recorded physiotherapy sessions, as quantification of the contents of the physiotherapy sessions would allow for process evaluation. The process evaluation of the physiotherapy actions revealed that some characteristics of COPCA were associated with improved developmental outcome. For example, in children at 18 months diagnosed with CP, i) the time spent on the physiotherapy action “challenging the infant to self-produced motor behaviour, continued by the infant with little variation”, had a positive association with the quality of the child's motor behaviour, and ii) the time spent on caregiver coaching had a positive correlation with the child's ability to adapt motor behaviour at 18 months CA. Other physiotherapy actions that were positively associated with the child's functional mobility were “family involvement and educational actions”, “postural support at the verge of the infant's abilities” and “challenging the infant to self-produced motor behaviour, continued by the infant with large variation”. In addition, the analyses indicated that spending more time on some TIP actions, such as handling techniques, was associated with worse developmental outcome [9,10]. In children with CP, the time spent on sensory experiences showed a negative correlation with the quality of the child's motor behaviour, and passive motor experiences were negatively associated with a neurological optimality score. In children without CP, more time spent on facilitation was associated with a lower functional mobility, and the time spent on “instructing the caregiver by means of assigning” showed a negative correlation with movement fluency at 18 months CA.

The aim of the present VIP follow-up study was to evaluate the effect of COPCA and TIP on outcome at school age. Long-term evaluation of early intervention is needed, as a) new associations between

physiotherapy actions during early intervention and outcome may emerge, as i) parents may continue to apply throughout childhood the physiotherapy principles they learned during early intervention, and ii) the child develops new functions that may depend on early life experiences; b) previously present associations between physiotherapy actions and outcome during infancy may fade and disappear. In line with the framework of the International Classification of Functioning Disability and Health, Child and Youth version (ICF-CY) and in accordance with the current focus of paediatric rehabilitation on activities and participation [11–13], we evaluated the children's functional outcome with assessment tools addressing the activity and participation domain. To this end, we used parental interviews and questionnaires to obtain information on the children's functional performance in daily life activities. Therefore, our primary outcome measurement was the Vineland Adaptive Behaviour Scale (VABS) [14]. Secondary outcome measurements included 1) the children's mobility and behaviour, and 2) parental coping strategies and educational approach. Next to the analyses of outcome at RCT level – and in line with the analyses performed for outcomes in infancy – we performed process evaluation in order to gain more insight into the possible working mechanisms of early intervention. We hypothesised that if families had incorporated the early intervention strategies into daily life, the earlier found associations between physiotherapy actions and developmental outcome might still be present.

## 2. Methods

### 2.1. Design overview

The present study is the follow-up of the VIP project, in which we compare functional outcome at school age of children who received either COPCA or TIP as early intervention. We sent an invitation letter to the parents of the VIP-children who participated in the final assessment of the original VIP-study at 18 months CA ( $n = 44$ , Fig. 1). The Medical Ethics Committee of the University Medical Centre Groningen approved the follow-up study (trial number NL39954.042.12).

### 2.2. Setting and participants

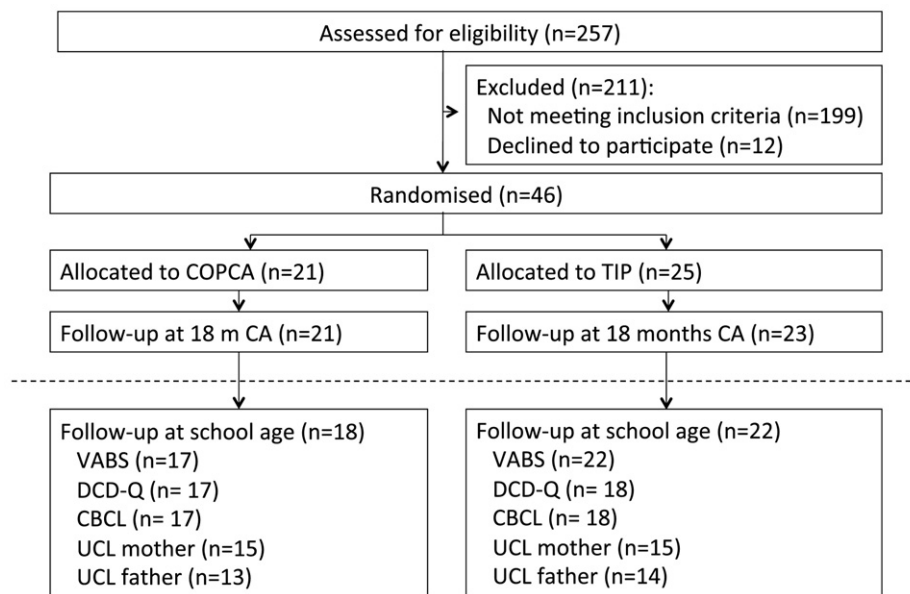
Inclusion in the VIP project was based on the presence of definitely abnormal general movements around 10 weeks CA (for details see references [9,10]). Infants with congenital anomalies and infants whose caregivers had an inappropriate understanding of the Dutch language were excluded. The infants had been admitted to the Neonatal Intensive Care Unit of the University Medical Centre Groningen between March 2003 and May 2005.

### 2.3. Randomization and interventions

Off-site participants had been randomly assigned with a random sequence generator to COPCA ( $n = 21$ ) or TIP ( $n = 25$ ). Intervention was applied between 3 and 6 months CA. The COPCA intervention took place in the home situation twice a week for 1 h. The frequency and location of the TIP intervention depended on the paediatrician's advice – it was mostly provided at home. Three infants assigned to TIP intervention did not receive physiotherapy. After the age of 6 months CA, the child's paediatrician decided whether to continue intervention [10]. Both COPCA and TIP interventions were provided by paediatric physiotherapists with over five years of experience in treating infants and children with special needs. A short description of the intervention in the two arms is provided below, for detailed information on the content of the interventions, see Dirks et al. and Blauw-Hospers et al. [5,9].

#### 2.3.1. COPCA

COPCA is a family relationship oriented programme that theoretically consists of two main components. The first component includes



**Fig. 1.** Flow diagram VIP project. For information on the original study design, infant recruitment and outcome at 18 months CA, see references [9,10]. The current study presents data on outcome at school age, i.e., the outcomes displayed below the dotted line. Abbreviations used: COPCA Coping with and Caring for infants with special needs; TIP Traditional Infant Physical therapy; CA corrected age; VABS Vineland Adaptive Behaviour Scale; DCD-Q Developmental Coordination Disorder Questionnaire; CBCL Child Behaviour Checklist; UCL Utrechtse Coping List.

family involvement and educational parenting. Development is considered a continuous dynamic interplay between child behaviour and family responses – along with environmental factors, which can influence both. Within COPCA, caregivers are coached to interact with their infant in order to respond to the infant's needs. As autonomy of the family is considered a key factor, the COPCA coach supports the family in defining priorities for intervention, in developing their own strategies of caring for their infants, and in improving personal coping skills.

The second component addresses neurodevelopment according to the principles of the NGST [7,8]. Development is considered a complex interaction between genetics and environmental influences, and is further characterized by two stages of variability. During primary variability, infants explore all variations within their motor repertoire. In the following (called: secondary) variability phase, the infant gradually learns to select the most efficient motor strategy for each situation. COPCA aims to challenge the infant to explore motor behaviour, and to promote variation in self-produced activities and trial-and-error experiences by means of play ('hands-off'), in order to let the infant explore his repertoire and learn to adapt his motor behaviour to the specifics of the situation.

COPCA coaches were educated via a two-day course, followed by four one-day sessions with an interval of six weeks. During the study, COPCA coaches continued to receive supervision from TD.

### 2.3.2. TIP

TIP in the Netherlands was at that time mostly based on the principles of neurodevelopmental treatment (NDT) [5,15]. Keywords of NDT are exposure to sensorimotor experience, facilitation of typical motor behaviour, and hands-on techniques. With time, influences of a more functional approach were incorporated into the treatment [15]. During the VIP project, TIP-therapists were frequently seen as a teacher or trainer [5]. Due to the eclectic approach of therapists, the application of NDT in daily practice is characterized by heterogeneity. The heterogeneity in TIP is associated with partial overlap in contents with COPCA. Nevertheless, TIP largely differs from COPCA, for example by spending virtually no time on coaching and substantially less time on challenging the infant to self-initiated activities than COPCA [5].

### 2.4. Quantification of contents of physiotherapy sessions in infancy

For the process evaluation, the actual content of the early intervention sessions was quantified. To this end, early intervention sessions in infants receiving either type of intervention had been video recorded at 4 and 6 months CA and analysed using the Observer software (Noldus, Wageningen, the Netherlands). The relative time spent on specific actions (labelled 'physiotherapy actions' as physiotherapists were the acting persons; Appendix A) was classified using the Groningen Observer Protocol. To allow comparison with future studies we re-analysed the physiotherapy videos with a new version of the Groningen Observer Protocol, i.e., version 2 (Hielkema T, personal communication 2016). In the new version two additional therapeutic approaches were included, i.e., actions typical for Vojta therapy and those typical for constrained induced movement therapy, to allow for an up to date and international application of the protocol. In addition, the category Self Produced Motor Behaviour was better specified. The new protocol has an excellent interrater reliability (median ICC main categories 0.945, range 0.677–0.998). For details on the original protocol, see references [5,9,10]. In line with the previous studies, we used the average time spent on physiotherapy actions observed at 4 and 6 months CA [9,10]. The video's were analysed by SJH and RJT, who were unaware of the infant's allocated intervention or outcome.

### 2.5. Outcomes at school age

The follow-up at school age (i.e., between 7.5 and 10 years) consisted of a parental interview (VABS) and parental questionnaires (Developmental Coordination Disorder Questionnaire [DCD-Q], Child Behaviour Check List [CBCL], the Utrechtse Coping List [UCL], and three questions on educational approach). The parental questionnaires were sent by surface mail to the family's home.

Our primary outcome measurement was the VABS, which was used to assess the children's functional performance in daily life [14]. The VABS is a scoring list that assesses by means of a structured interview the functional status in communication, daily living skills, socialization and motor skills. For each VABS item, a score 2 (yes, the child performs this action in daily life), score 1 (the child is sometimes or partly capable

of performing this action), or score 0 (the child never performs this action) is assigned by the parents. Examples of functional items are: 'can use utensils', 'knows the rules at home and at school', 'has friends', 'can use a bike'. This means that the VABS addresses the ICF-domains of activity and participation. The VABS items are categorized into four domains that are further divided in eleven subdomains, for example receptive communication and fine motor skills. The VABS can be applied from birth to adulthood, with exception of the motor domain that can only be used until the age of 6 years in typically developing children. The VABS has a good reliability and validity in both typically developing children and in children with developmental disorders, such as CP [16, 17]. The VABS was carried out by EGH, as part of a telephone interview in which also information on the child's medical history and education was obtained. In the Netherlands children with clear learning disabilities, behavioural problems, or physical problems, including deafness or blindness often attend a school for special education. EGH was blinded to all child and family characteristics (including type of intervention), except for the child's sex and age.

As the VABS has age limitations in the motor domain, we added the DCD-Q. The DCD-Q is a brief parental questionnaire designed to identify motor problems in children, which may indicate the presence of Developmental Coordination Disorder (DCD) [18]. The DCD-Q may also be used as a questionnaire to evaluate mobility in children with and without CP [19,20]. The DCD-Q evaluates performance of motor skills including control during movement, fine motor skills/handwriting, gross motor skills, and general coordination. The DCD-Q total score was used to assess mobility, since the majority of items can be classified within the mobility domain of the ICF-CY (e.g., 'your child jumps easily over obstacles found in garden or play environment'; d4553). The DCD-Q is a reliable and valid instrument [21], as is the Dutch translation [22].

The child's behaviour was assessed with the CBCL; a widely used parental questionnaire containing 113 items [23]. We calculated the internalizing, externalizing and total scores. In addition, scores of the Diagnostic and Statistical Manual of Mental Disorders (DSM) oriented scales were calculated and dichotomized as typical versus atypical (borderline and clinical range). The Dutch CBCL has a good reliability and validity [23].

Both parents were asked to complete the UCL. The UCL evaluates a person's ability to cope with stressful situations and was used to document parental coping [24]. The questionnaire consists of 47 items, scored on a four-point scale. The seven subscales were calculated such as active tackling or passive reacting. The UCL has a satisfactory reliability and validity [25].

Lastly, parents received a form with three study specific fixed-choice questions on their educational approach: 1) if they still applied the principles which they had learned during early intervention, 2) if the early intervention had influenced their educational approach, and 3) their approach of the child when he/she learns new skills (for the full text of the questions see Appendix B).

## 2.6. Statistical analysis

The sample size of the original VIP-project was based on the outcome on the Infant Motor Profile at the age of 18 months [10]. The present follow-up study allowed for a detection of a difference in VABS score of 14 points with a power of 80% ( $\alpha = 0.05$ ,  $SD = 15$ ). Based on the results of the follow-up at 18 month we expected to find at RCT-level minor differences at best. Therefore, we had planned to perform in addition to the RCT-analysis a process evaluation.

Statistical analysis was performed using SPSS software version 20. Possible differences between COPCA and TIP in dichotomized outcome parameters (e.g. attendance of special education) were investigated using the Fisher exact test and  $\chi^2$  test and expressed as Odds Ratios (OR) with 95% confidence intervals. The Mann-Whitney *U* test was performed to investigate differences in continuous outcome measurements, e.g., VABS scores. Analysis followed the principles of

intention to treat. Differences with a *p*-value  $<0.05$  were considered to be statistically significant.

For the process evaluation partial correlations were used to correlate the time spent on physiotherapy actions (neuromotor actions, educational actions and communication) with DCD-Q, CBCL and UCL scores, using two control variables (presence of CP and maternal education). As VABS scores typically depend on the child's age, an additional control variable (age at follow-up) was used in the correlational analyses between physiotherapy actions and VABS scores. Only correlations with a *p*-value  $<0.01$  were considered statistically significant to correct for multiple testing. For the process evaluation of the association between dichotomized parental educational outcome parameters and physical therapy actions the data were explored with the Mann-Whitney *U* Test.

## 3. Results

Parents of 40 children (91%, median age 8 years and 4 months, 20 girls, 18 COPCA) participated in the follow-up study (Tables 1 and 2). The responders and non-responders did not differ in baseline characteristics (sex, gestational age, birth weight, educational level of the parents, maternal age at delivery, type of intervention and prevalence of CP at 18 months CA - data not shown). Also the two intervention groups did not differ significantly in baseline characteristics (Table 1). In our group of infants at high risk for CP, gestational age at birth was not associated with the VABS, DCD-Q and CBCL scores.

### 3.1. Outcome at the level of the RCT

On RCT-level, outcome at school age of children who had received COPCA was similar to outcome of children who had received TIP in terms of diagnosis of CP, attendance of special education, use of additional paramedical therapies, VABS- (including subdomain scores - not shown), DCD-Q- and CBCL scores (Table 2). Two infants assigned to TIP had not received early intervention [9,10]; their development did not differ from the development of the other TIP children (data not shown).

**Table 1**  
Participants: characteristics.

	COPCA ( <i>n</i> = 18)	TIP ( <i>n</i> = 22)	<i>p</i> -Value
Sex, n (male/female)	9/9	11/11	1.000
GA in weeks, median (range)	30 (27–40)	30 (26–39)	0.744
Term born	2	2	
Between 28 and 36 weeks GA	12	16	
Below 28 weeks GA	4	4	
BW in grams, median (range)	1415 (670–4750)	1205 (635–3460)	0.254
Member of set of twins, n	8	6	0.327
Firstborn child, n	8	9	1.000
Maternal age at child birth in years, median (range)	31 (21–43)	32 (22–39)	0.591
Severe brain lesion <sup>a</sup> , n	2	3	1.000
Maternal education, n			
Low	3	3	0.111
Middle	13	10	
High	2	9	
Paternal education, n			
Low	9	6	0.311
Middle	6	11	
High	3	4	
Siblings, n			
None	2	3	0.539
One	11	12	
Two or three	5	7	
Divorced parents, n	2	4	0.679

<sup>a</sup> IVH grade 4 or PVL grade 3–4.

**Table 2**  
Outcome at school age.

	COPCA ( <i>n</i> = 18)	TIP ( <i>n</i> = 22)	<i>p</i> -Value	OR (95% CI)
Age at follow-up in years + months, median (range)	8 + 6 (7 + 6–10 + 1)	8 + 2 (7 + 5–9 + 6)	0.788	n.a.
Cerebral palsy, <i>n</i> <sup>a</sup>	2	4	0.673	1.8 (0.3–11.0)
Special education, <i>n</i> <sup>b</sup>	6	4	0.300	0.4 (0.1–1.9)
VABS score <sup>c</sup> , median (range)				
Communication	230 (185–239)	231 (193–246)	0.599	n.a.
Daily competences	271 (51–285)	267 (134–314)	0.966	n.a.
Socialization	171 (113–181)	166 (136–184)	0.533	n.a.
Locomotive skills	144 (15–146)	144 (67–146)	0.647	n.a.
DCD-Q score <sup>d</sup> , median (range)	65 (33–78)	63 (25–85)	0.503	n.a.
Suspected + borderline DCD-Q score, <i>n</i>	3 + 1	4 + 1	1.000	
CBCL <sup>e</sup> , median (range)				
Internalizing score	4 (0–32)	7 (2–24)	0.163	n.a.
Externalizing score	4 (0–22)	2 (0–12)	0.287	n.a.
Total score	18 (0–68)	23 (5–65)	0.909	n.a.
Clinically relevant problems, <i>n</i> (DSM scale)	4	5	1.000	1.3 (0.3–5.7)
Additional therapies, <i>n</i> (total)	11	14	0.750	1.3 (0.3–4.7)
Physical therapy	5	10	0.323	2.4 (0.6–9.0)
Speech therapy	8	8	0.746	0.8 (0.2–2.8)
Occupational therapy	1	4	0.348	4.0 (0.4–39.8)
UCL scores, median (range)				
Active tackling - M	17 (11–25)	19 (15–22)	0.512	n.a.
Seeking social support - M	15 (10–20)	17 (11–24)	0.106	n.a.
Palliative reacting - M	16 (10–17)	15 (12–21)	0.412	n.a.
Avoiding - M	12 (6–20)	15 (11–19)	0.106	n.a.
Passive reacting - M	9 (7–15)	11 (8–19)	0.325	n.a.
Reassuring thoughts - M	6 (4–9)	6 (4–8)	0.461	n.a.
Expression of emotions - M	12 (8–15)	11 (8–20)	0.870	n.a.
Active tackling - F	20 (11–24)	18 (12–25)	0.488	n.a.
Seeking social support - F	14 (8–22)	17 (9–21)	0.169	n.a.
Palliative reacting - F	14 (8–17)	16 (9–20)	0.325	n.a.
Avoiding - F	10 (6–16)	11 (7–17)	0.280	n.a.
Passive reacting - F	9 (7–18)	11 (7–19)	0.430	n.a.
Reassuring thoughts - F	6 (3–9)	6 (3–9)	0.867	n.a.
Expression of emotions - F	11 (5–14)	11 (7–15)	0.550	n.a.

VABS Vineland Adaptive Behaviour Scale; DCD-Q Developmental Coordination Disorder Questionnaire; CBCL Child Behaviour Checklist; UCL Utrechts Coping List; n.a. not applicable; M = mother, *n* = 30; F = father, *n* = 27.

<sup>a</sup> Three children with CP were independent walkers, all of them had received TIP.

<sup>b</sup> Six of the ten children who received special education had CP, one had speech problems, three children had no specific medical diagnosis nevertheless they could not cope with main stream education.

<sup>c</sup> *n* = 39, see Fig. 1; 4 of the 9 children with a CBCL-score in the clinical range attended a school for special education.

<sup>d</sup> *n* = 35, see Fig. 1; 5 of the 9 children with a DCD-score suspect for DCD attended a school for special education.

Also parental UCL scores in the two groups did not differ (Table 2). Parents of 35 children filled out the questionnaire with regard to educational approach. Four parents in the COPCA group and two parents in the TIP group mentioned that they still applied the principles they had learned during early intervention (Fisher's Exact Test; *p* = 0.402, OR 2.5 [0.4–15.6]). Six parents in the COPCA and five parents in the TIP group reported that the early intervention had influenced their educational approach (Fisher's Exact test; *p* = 0.725, OR 1.4 [0.3–5.9]). Parental approach when the child learns new skills differed between the two intervention groups: parents of six children of the COPCA group and one of the TIP group indicated that they let their child try until it succeeds (with trial and error) (Fisher's Exact test; *p* = 0.041, OR 9.3 [1.0–87.9]). The other parents (*n* = 28) reported that they first let their child try and start to intervene when the child seems unsuccessful.

### 3.2. Process evaluation

Videos of early intervention sessions were available for analysis for 38 out of the 40 participating children. Two physiotherapy actions were associated with developmental outcome at school age. The actions belong to the domains of education and communication. First, the time spent on caregiver training had a negative correlation with mobility (DCD-Q; *r* = −0.511, *p* = 0.003). Caregiver training was defined as all actions during which the physiotherapist instructs the caregivers, for instance how to present a toy, or how to use specific techniques, e.g. by demonstrating handling techniques. Second, the total time spent on instruction of the caregiver, defined as all communication in

which the caregiver is given assignments, hints or strict directions regarding treatment strategies, showed an inverse association with mobility (DCD-Q; *r* = −0.514, *p* = 0.003). This association could be attributed especially to the provision of strict instructions: the time spent on strict instructions on the best way to perform had an inverse relationship with mobility (*r* = −0.478, *p* = 0.006). The reported correlations did not depend on the child being in special education or not. None of the neuromotor actions, such as the time spent with facilitation or the time spent with being challenged to self-produced motor behaviour (SPMB), were associated with child outcome at school age.

None of the physiotherapy actions were associated with parental coping strategies (UCL-scores). The exploratory process evaluation of the associations between physiotherapy actions and parental educational strategies revealed the following. Parental report of the application of principles they had learned during early intervention (Appendix B, question 1) was not associated with specific physiotherapy actions. However, parents who reported that the early intervention had influenced their educational approach (question 2) had received more feedback during the early intervention sessions (19% vs. 12% of intervention time, Mann-Whitney *U*; *p* = 0.008, Fig. 2a). In addition, in families who indicated that they let their child try until it succeeds while learning a new skill (with trial and error, question 3), more time was spent on caregiver coaching (74% vs. 0% of intervention time, Mann-Whitney *U*; *p* = 0.004) and on SPMB of the infant (50% vs. 41% of intervention time, Mann-Whitney *U*; *p* = 0.021), and less time was spent on facilitation techniques (0% vs. 7% of intervention time, Mann-Whitney *U*; *p* = 0.005) during the early intervention, compared to

families who indicated that they first let their child try and start to intervene when the child seems to be unsuccessful (Fig. 2b–d). Parental educational strategies, as measured by our study-specific fixed choice questionnaire, were not related to the child's VABS, DCD-Q or CBCL score or to the educational level of the parents. However, parents of children with CP more often applied the principles they had learned during early intervention than parents of children without CP (CP 60% vs. no-CP 10%, Chi-square;  $p = 0.026$ ). A similar effect was found for parents of all children attending a school for special education: they applied the principles they had learned during early intervention more often than parents of children attending mainstream education (44% vs. 8%, Chi-square;  $p = 0.027$ ).

#### 4. Discussion

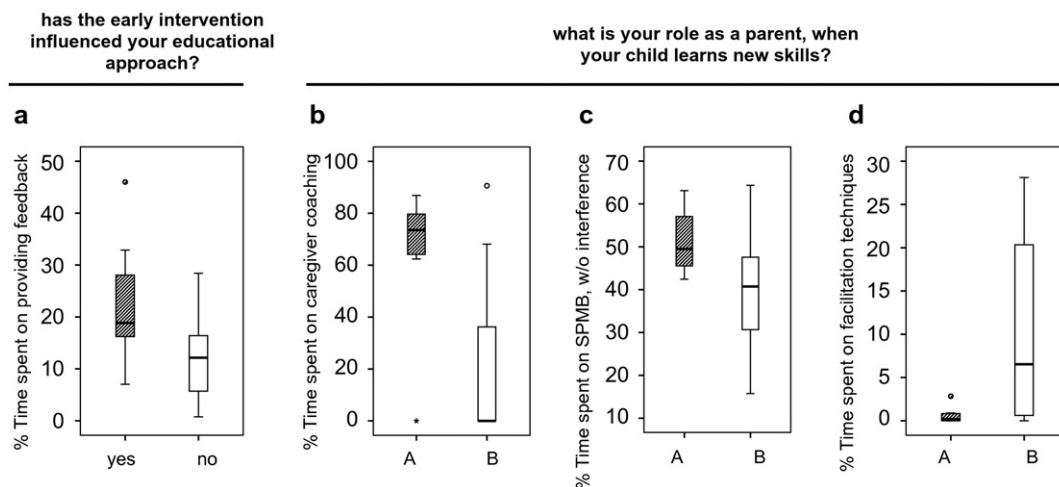
On RCT-level, functional outcome at school age of children who had received 3 months of COPCA was similar to outcome of children who had received TIP as early intervention. Yet, the RCT revealed a minor difference in parental educational approach between the groups. Parents whose infant had received the COPCA intervention more often used a trial and error approach when the child learned a new skill than parents of children who had received TIP. In addition, the process evaluation revealed that two physiotherapy actions were associated with child mobility and that four other physiotherapy actions were related to parental educational approach.

Our RCT-results are in line with the few studies on long-term effects of early intervention, suggesting that the effect of early intervention does not extend to school-age [1]. However, we previously argued that the RCT design in fact may be not the best way to unravel effectiveness of early physiotherapy intervention, as the various interventions are heterogeneous and partially overlap [5,9]. The process analyses allowed us to explore which physiotherapy actions were associated with outcome at school age. We found that two physiotherapy actions were associated with DCD-Q scores: more time spent on caregiver training and strict instructions was associated with worse mobility at school age. These findings are in line with theories on active learning. They emphasize the importance of self-directed learning and the development of problem solving skills, with the professional acting as a coach or consultant, rather than telling (training) the parents or child what to do in a specific situation [26,27].

Four physiotherapy actions were associated with parental reports on educational approach. Families, where more time had been spent on the provision of feedback, i.e., an active communication style in which caregiver and therapist evaluate the intervention or share information, later reported that early intervention continued to influence their educational approach. Three other actions were associated with the parents' approach of the child when he/she learns new skills. Parents who reported that they let their children learn by trial-and-error, i.e., who used neurodevelopmental principles of the COPCA programme, had received more coaching, and during intervention their child had spent more time with self-produced motor behaviour. Both coaching and letting the child perform motor actions without adult assistance, are key elements of COPCA. Indeed, the parents in the COPCA group more often reported the trial-and-error approach than the parents in the TIP group. On the contrary, in families who indicated at follow-up that they start to intervene when the child seems to be unsuccessful, more physiotherapy time was spent on facilitation, a TIP-action based on NDT [15]. We realise that the associations between the content of early intervention and parental behaviour are weak. Nevertheless, it is possible that such long-term effects may exist.

Our results partly correspond to the previously reported associations between physiotherapy actions and outcome at 18 months CA (see references [9,10]). At that time, subgroup analyses for children with and without CP were performed. The limited number of children with CP in the current follow-up precluded subgroup analyses. In line with results on outcome at school age, previous analysis revealed 1) a positive association between the time spent on caregiver coaching (i.e., the opposite of caregiver training) and motor outcome (spontaneous movement quality, subdomain variation) in infants with CP and 2) a negative association between the time spent on instruction of the caregivers and movement fluency in children without CP. In contrast to the previous study, we could not demonstrate an association between early intervention time spent on neuromotor actions and outcome at school age. This implies that previously existing associations had faded, e.g., the association between “challenging the infant to self-produced motor behaviour” and better motor scores, and the associations between the neuromotor actions facilitation, sensory experience and passive motor experience and worse motor outcome at 18 months.

To summarize, the VIP project demonstrated associations between the professional approach of the physiotherapist ('coach' vs. 'instructor') and long-term outcome: less training and less instruction were



**Fig. 2.** Educational approach and physical therapy actions. The boxplots illustrate the percentage of time spent with a specific physical therapy action during early intervention in the subgroups organized according to the answers parents provided on the educational questionnaire (Appendix B). Panel a: Question 2, the question on educational approach: has the early intervention influenced your educational approach (yes [ $n = 11$ ] or no [ $n = 24$ ]). Panels b, c, d: Question 3, the question on parental approach when the child learns new skills: A) they let their child try until it succeeds (with trial and error) ( $n = 7$ ), B) they first let their child try and start to intervene when the child seems unsuccessful ( $n = 28$ ), C) they prefer to assist their child in learning new skills in order to avoid that the child is confronted with difficulties (option not chosen by any of the parents). Data are presented as median values (horizontal bars), interquartile ranges (boxes) and ranges (vertical lines) with outliers (circles). SPMB = Self Produced Motor Behaviour.

associated with better outcome in the mobility domain – which is a major goal of COPCA. We could not demonstrate long-term effects of specific neuromotor actions during early intervention and long-term mobility.

The strengths of the present study are the long-term follow-up and the detailed process evaluation of the early intervention by means of video-analysis. The presence of both heterogeneity and overlap in early intervention [5] may explain why we only found associations between physiotherapy actions and outcome, but did not find differences on RCT-level. Our VIP project stresses the importance of process evaluation and underscores the notion that a strict RCT design is often not realistic in studies on early intervention [9,10], as there is both overlap and heterogeneity in early intervention programmes [1,2,9,10]. Yet, we would like to emphasize that correlations are not identical with causations. Another limitation is the small sample size: it allowed only for the detection of relatively large differences at RCT-level. The use of parental questionnaires to assess developmental outcome also could be considered a limitation as they provide subjective views of parents and not a professional assessment. However, we deliberately used these assessment tools as 1) our focus was on the activity and participation domain of the ICY-CY, and 2) parents are pre-eminently able to provide information on functional outcome in daily life. In addition, we consider the ceiling effect of the VABS motor skills domain as a limitation. This may explain why we did find associations between physiotherapy actions and mobility (measured with the DCD-Q), but not with functional motor skills (measured with the VABS). We do consider a gain of a few points on the DCD-Q questionnaire as relevant in daily life. Follow-up studies on early intervention are typically hampered by heterogeneity in type and frequency of additional therapies that started after the early intervention period and other environmental factors. In the present study, the number of children that did receive additional paramedical therapies was similar in both groups (Table 2). Finally, the intervention period of 3 months was short.

#### 4.1. Conclusions

In conclusion, this study indicates that functional outcome at school age of infants who received three months of COPCA and that of infants who received three months of TIP is similar. However, some specific physiotherapy actions were associated with the children's mobility and parental educational approach at follow-up. Apparently, the professional approach of the physiotherapist can make a difference.

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#### Appendix A. Physical therapy actions during early intervention

##### Treatment situation

- Motor activity/play
- Feeding
- Bathing

- Dressing/Undressing
- Changing Diapers
- Carrying

##### Neuromotor actions

- Facilitation techniques
- Sensory experience
- Passive motor experience
- Self-produced motor behaviour (SPMB), no interference with physical therapist (PT) or caregiver
- Challenged to SPMB (CSPMB), infant is allowed to continue activity by him/herself
- CSPMB, activity flows over into facilitation, sensory or passive experience

##### Educational actions toward caregiver

- Caregiver training
- Caregiver coaching

##### Communication between PT and caregiver

- Information exchange
  - Regarding family history, NICU experiences, current situation or daily business
  - Regarding principles of NDT
  - Regarding principles of COPCA
- Instruct
  - PT gives strict instruction
  - PT provides multiple options
  - PT provides hints
- Provide feedback
  - PT and caregiver share information
  - PT asks and listens to the opinion of the caregiver
  - PT evaluates the procedure
  - PT tells the caregiver what went right or wrong
- No communication

For additional information see references [5,9,10].

#### Appendix B. Questions on educational approach (translated from Dutch into English)

Questions 1: do you still apply the principles that you have learned during early intervention?

- a. yes
- b. no

Question 2: has the early intervention influenced your educational approach?

- a. yes
- b. no

Question 3: what is your role as a parent, when your child learns new skills?

- a. I let my child try until he/she succeeds (with trial and error)
- b. I first let my child try and start to intervene when the child seems unsuccessful
- c. I prefer to assist my child in learning new skills in order to avoid that the child is confronted with difficulties



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