



## REGULAR ARTICLE

# The Coping with and Caring for Infants with Special Needs intervention was associated with improved motor development in preterm infants

Schirin Akhbari Ziegler<sup>1</sup>  | Michael von Rhein<sup>2,3</sup> | André Meichtry<sup>1</sup> | Markus Wirz<sup>1</sup> | Tjitske Hielkema<sup>4</sup> | Mijna Hadders-Algra<sup>5</sup>  | the Swiss Neonatal Network & Follow-Up Group

<sup>1</sup>School of Health Professions, Institute of Physiotherapy, Zurich University of Applied Sciences ZHAW, Winterthur, Switzerland

<sup>2</sup>Department of Pediatrics, Kantonsspital Winterthur, Winterthur, Switzerland

<sup>3</sup>University Children's Hospital Zurich, Zurich, Switzerland

<sup>4</sup>Department of Pediatrics, Division of Developmental Neurology, University Medical Center Groningen, University of Groningen, Groningen, The Netherlands

<sup>5</sup>University of Groningen, University Medical Center Groningen, Department of Pediatrics, Division of Developmental Neurology, Groningen, The Netherlands

## Correspondence

Schirin Akhbari Ziegler, Zurich University of Applied Sciences School of Health Professions, Institute of Physiotherapy, Katharina-Sulzer-Platz 9, 8401 Winterthur, Switzerland.  
Email: akhb@zhaw.ch

## Funding information

This study was supported by the Schweizerische Stiftung für das cerebrally gelähmte Kind, the Anna Mueller Grocholski Foundation, the Jubiläumsstiftung der Schweizerischen Mobiliar Genossenschaft and the research fund of the Swiss Association of Physiotherapy.

## Abstract

**Aim:** We compared the impact of standard infant physiotherapy and the family-centred programme, Coping with and Caring for Infants with Special Needs (COPCA), in infants born before 32 weeks without significant brain lesions.

**Methods:** This randomised controlled trial was carried out in patients' homes and outpatient settings in Switzerland between January 2016 and October 2019. We used data from the national SwissNeoNet register and an assessment battery that included infant and family outcomes and video analyses of therapy sessions. The Infant Motor Profile was the primary outcome instrument.

**Results:** The COPCA group comprised six boys and two girls with a median gestational age of 27 weeks (range 25-30), and the standard care group comprised seven boys and one girl with a median gestational age of 29.5 weeks (range 26-31). COPCA participants improved significantly more between baseline and 18 months in the IMP variation (9.0 percentage points, 95% confidence interval: 0.3-17.5) and performance (12.0 percentage points, 95% confidence interval: 4.1-20.6) domains than standard care participants. COPCA coaching was positively associated with IMP scores at 18 months, but some standard care actions were negatively associated.

**Conclusion:** COPCA was associated with better motor outcome in infants born before 32 weeks than standard infant physiotherapy.

## KEYWORDS

early physiotherapy, family-centred practice, family outcomes, motor outcome, preterm infants

**Abbreviations:** BSID-III, Bayley Scales of Infant and Toddler Development, Third Edition; CI, confidence interval; COPCA, Coping with and Caring for Infants with Special Needs; IMP, Infant Motor Profile; PEDI, Pediatric Evaluation of Disability Inventory; RCT, randomised controlled trial.

This is an open access article under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made.

©2020 The Authors. *Acta Paediatrica* published by John Wiley & Sons Ltd on behalf of Foundation Acta Paediatrica

## 1 | INTRODUCTION

Despite improvements in perinatal and neonatal care, infants born before 32 weeks of gestation face a substantially increased risk of motor and cognitive impairments. About 5%-9% of these preterm infants develop cerebral palsy.<sup>1</sup> Many more infants are diagnosed with milder disorders later in life, including mild cognitive, motor impairment and attention-deficit/hyperactivity disorder.<sup>1,2</sup>

In Switzerland, all children born below 32 weeks of gestational age are assessed for developmental outcomes and the need for therapeutic support in specialised follow-up centres, according to a standardised programme. Outcome data are entered into the national SwissNeoNet register.<sup>3</sup> This register is run by the Swiss Society of Neonatology and is a medical quality register for level 3 and level 2b units. Participation by these units is mandatory under Swiss law.

Various early physiotherapeutic intervention programmes are available for preterm born children with motor impairments.<sup>4</sup> Unfortunately, no evidence is available about whether physiotherapeutic intervention programmes for infants have a positive effect on long-term motor outcomes.<sup>4</sup> Until about two decades ago, traditional physiotherapeutic approaches focused on the child's motor development. Since then, physiotherapeutic intervention programmes have increasingly included caregivers. Family-centred care, which recognises the importance of including family members as active and equal partners in the child's care, has become the practice of choice.<sup>5</sup> Also, the focus on early physiotherapeutic interventions has shifted to the parents when preterm infants are born before 32 weeks. Systematic reviews on early interventions have indicated that parental involvement was associated with better outcomes for both the infant and family.<sup>4,5</sup>

Coping with and Caring for Infants with Special Needs (COPCA) is an early physiotherapeutic intervention family-centred programme, which was developed some 15 years ago.<sup>6</sup> It comprises a family and educational component and a neurodevelopmental component. The family component focuses on including family members as autonomous, responsible and active partners in the intervention process. The coach, who is a physiotherapist, encourages the family to stimulate the infant's development during daily care and to make their own decisions. The educational component is based on neuronal group selection theory, which is a framework for understanding and treating disordered motor behaviour in children. This component challenges the infant's self-produced motor behaviour with trial and error experiences. It aims to increase the variety of the infant's motor movements and to enhance the infant's capacity to adapt movements to different situations.<sup>7</sup>

Studies from the Netherlands compared the effect of the COPCA intervention programme with standard infant physiotherapy care, which is referred to as standard care in this paper.<sup>6,8</sup> Both studies evaluated COPCA in infants at very high risk of cerebral palsy and used a double approach, by combining a randomised controlled trial (RCT) with process evaluation. The latter<sup>8</sup> included video recordings of the intervention sessions, in which distinguishable intervention elements were defined, quantified and analysed with respect to

### Key notes

- The Coping with and Caring for Infants with Special Needs (COPCA) intervention had not been evaluated in preterm infants born at less than 32 weeks of gestation without significant brain lesions.
- Improvements in the Infant Motor Profile were the primary outcome instrument in this study, with a particular on the variation and performance domains.
- COPCA was associated with better motor outcomes in our cohort of 16 infants than standard infant physiotherapy.

their association with developmental outcomes.<sup>6,9-11</sup> The first study<sup>6</sup> comprised 46 term and preterm infants who had definitely abnormal general movements at 10 weeks of corrected age. Of these, 10 (22%) were later diagnosed with cerebral palsy. The infants received either COPCA or standard care between 3 and 6 months of corrected age, and the authors reported that the developmental outcomes of the two intervention groups did not differ. This could partly be explained by the overlap in physiotherapeutic content between the two interventions. The process evaluation showed that the associations between the contents of the physiotherapy and developmental outcomes differed for children who were, and were not, diagnosed with cerebral palsy. In the infants with cerebral palsy, two important COPCA components were associated with better motor developmental outcomes at 18 months of corrected age: caregiver coaching and challenging the infant to self-produce motor behaviour with ample trial and error experiences.<sup>6,10</sup> In children without cerebral palsy, the use of hands-on techniques was associated with less favourable motor outcomes.<sup>6</sup> The second study comprised 43 term and preterm infants at very high risk of cerebral palsy, mainly due to severe brain lesions. After they were included between term-equivalent age and 9 months of corrected age, the infants received COPCA or standard care for 12 months. The authors reported that 22 infants (51%) were later diagnosed with cerebral palsy.<sup>10,11</sup> Again, the groups did not differ in developmental outcomes and the content of the physiotherapy was not associated with developmental outcomes. However, a difference in family function emerged and that was that the family's quality of life improved over time in the COPCA group, whereas it remained similar in the standard care group. In addition, a qualitative study on caregivers' experiences with COPCA showed that the mothers of infants with special needs appreciated the intervention programme.<sup>12</sup> This was in line with another study that showed that more frequent caregiver coaching was associated with better family empowerment.<sup>11</sup> Receiving the intervention at home, support from the COPCA coach, the chance to participate as an active partner and the parents' increasing awareness of their ability to make independent, informed decisions were particularly valued.

However, the COPCA intervention programme had not been evaluated in the much larger group of preterm infants born before

32 weeks without significant brain lesions. Therefore, the primary aim of this small explorative study was to evaluate the effect of 6 months of the COPCA intervention programme. The programme focused on preterm infants born before 32 weeks of gestation and was applied from 1 to 4 months of corrected age onwards. These were infants who all faced a moderate-to-high, but not very high, risk of developmental disorders. The focus was on the infant's motor development, as the major aim of early physiotherapy interventions is to improve motor outcomes in infants with, or at risk of, developmental motor disorders. In addition, we aimed to evaluate the effects of the intervention on infant cognition and family outcomes. The double approach of an RCT and process evaluation was used, similar to the Dutch studies described above. We addressed four research questions. First, did motor development differ between preterm infants receiving COPCA and controls receiving standard care when it was measured immediately after the intervention and at 18 months of corrected age using the Infant Motor Profile (IMP)? Second, did cognitive development and function in daily life differ in the two intervention groups, immediately after the intervention and at 18-24 months of corrected age? Third, did family outcomes improve immediately after the intervention and at 18 months of corrected age, when infants received COPCA or standard care? Fourth, what were the specific physiotherapy actions associated with better motor outcomes at 18 months of corrected age?

## 2 | PATIENTS AND METHODS

This explorative multicentre, parallel-group study, with balanced randomisation, was conducted in the German-speaking part of Switzerland and stratified by its four study sites in Zurich, Winterthur, Basel and St. Gallen.

### 2.1 | Participants

The primary inclusion criterion was preterm birth at <32 weeks of gestation. The neonatal data on all infants were retrieved from the standard recording in the SwissNeoNet register.<sup>3</sup> The infants were recruited between 35 weeks of gestational age and 4 months of corrected age, if they showed neurological abnormalities that indicated a moderate-to-high risk of cerebral palsy. Neurological symptoms included muscular hypertonia, hypotonia, hyperexcitability and abnormal general movements or cranial ultrasound abnormalities. Referrals to paediatric physiotherapy by the physicians in the follow-up centres were based on abnormal neurological findings. The exclusion criteria were additional severe congenital disorders, such as a serious congenital heart disorder or cystic fibrosis. Infants who participated in the Erythropoietin for the Repair of Cerebral Injury in Very Preterm Infants study were excluded. Caregivers who had a poor understanding of German were also excluded.

Between 29th February 2016 and 4th December 2017, 22 infants seen at the four study sites fulfilled the inclusion criteria and 16 were included, as four families did not want to take part and two infants were seeing a therapist who was not participating in the study (Figure 1). After written, informed consent, the infants were randomly assigned to the two groups. The COPCA group comprised six boys and two girls with a median gestational age of 27 weeks (range 25-30) at birth and median age of 4.5 weeks (range 0-15) at inclusion. The standard care group consisted of seven boys and one girl born at a median of 29.5 weeks (range 26-31) and included at a median age of 14.5 weeks (range 0-22) (Table 1).

The computer generated a set of permuted blocks with different lengths of numbers of participants for each site. The random allocation sequences were sent to a study-specific mail account, from which the neonatologist could retrieve the infant's study group allocation. The infant was allocated to receive standard care or COPCA.

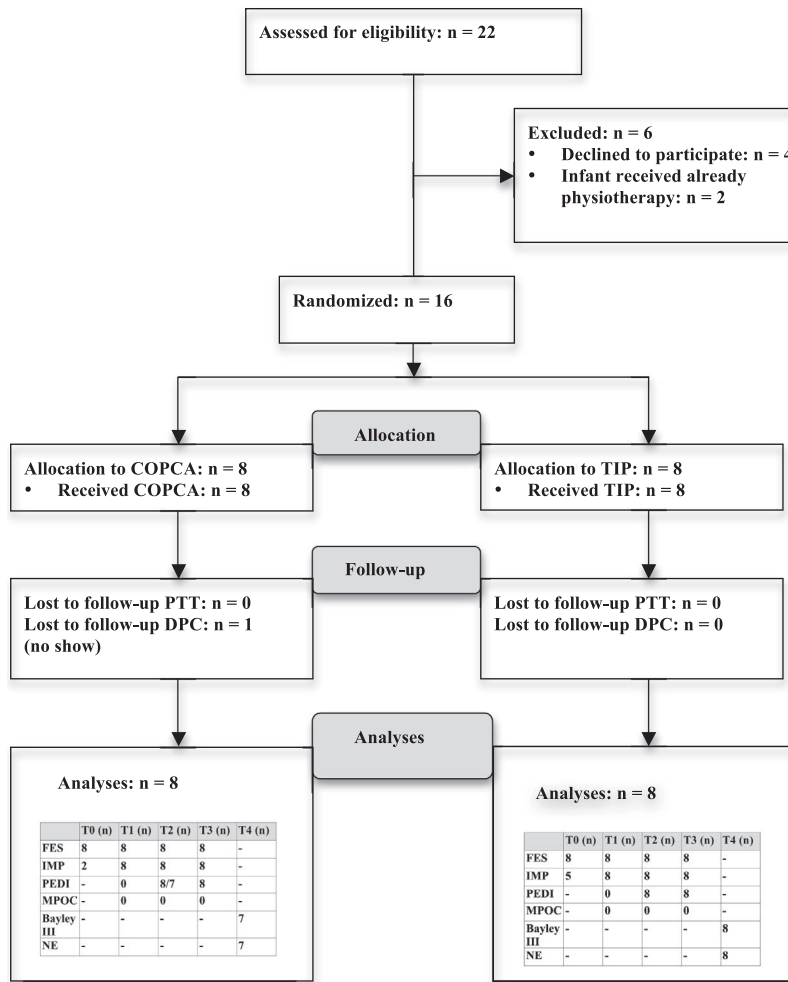
### 2.2 | Interventions

Participants received COPCA or standard care for 6 months after inclusion, with a recommended frequency of one weekly face-to-face session lasting 30-45 minutes. All eight of the COPCA families received the intervention at home. The standard care intervention was provided at home in two cases and in outpatient settings in six cases.

The intervention was delivered by COPCA coaches who followed the programme's theoretical and practical principles.<sup>13</sup> Caregivers learnt how to stimulate their infant's development by challenging their motor behaviour with trial and error experiences (Figure 2A). This aimed to empower the caregivers' competencies to stimulate the infant's daily development, by increasing their motor repertoire and enhancing their capacity to adapt movements to situations. Standard care was based on what paediatric physiotherapists generally assume to be useful to promote the development of infants with special needs. Standard care is heterogeneous and eclectic, uses parent training and often includes components of neurodevelopmental treatment with hands-on techniques (Figure 2B).<sup>6</sup> Details of both interventions are provided in Appendix S1, according to the Template for Intervention Description and Replication guidelines.<sup>13-15</sup>

### 2.3 | Measurements

The study's primary outcome measure was the IMP,<sup>16</sup> which was performed at baseline, 3 and 6 months after baseline and at 18 months of corrected age. The IMP is a video-based assessment that provides information on the infant's motor behaviour in five domains: variation or the size of the motor repertoire, adaptability or the ability to select adaptive motor strategies, symmetry, fluency and performance. The total score and domain scores are expressed as percentages, with a maximum score of 100%. The IMP has good reliability and validity, including proper responsiveness to change.<sup>9,17,18</sup> The



**FIGURE 1** Flow diagram of participants and enrolment. BSID-III: Bayley Scales of Infant and Toddler Development, Third Edition; COPCA, Coping with and Caring for Infants with Special Needs; DPC, Developmental Pediatric Center; FES, Family Empowerment Scale; IMP, Infant Motor Profile; MPOC, Measure of Processes of Care; NE, neurological examination; PEDI, Pediatric Evaluation of Disability Inventory; PPT, Pediatric Physiotherapy; SC, standard care

IMP assessment was carried out by the therapist in charge of the infant's intervention and the video was scored by an assessor who was blinded to the group allocation.

The secondary outcome measures were the Family Empowerment Scale,<sup>19</sup> the Pediatric Evaluation of Disability Inventory (PEDI),<sup>20,21</sup> the Measure of Processes of Care,<sup>22,23</sup> the Bayley Scales of Infant and Toddler Development, Third Edition (BSID-III)<sup>24</sup> and a neurological examination. The Family Empowerment Scale is a questionnaire that assesses empowerment of the family regarding family life, exposure to the child's health care service and the parents' community involvement. We only used the German translation of the 12-item family subscale, with a maximum total score of 60. The German version of the PEDI was used to assess functional outcome and assistance needs. The German version of the Measure of Processes of Care parental questionnaire was used to quantify the extent to which the caregivers experience family-centredness in child care.<sup>22,23</sup> The BSID-III<sup>24</sup> and the neurological examination were carried out by experienced developmental paediatricians, blinded to the group allocation, during the routine follow-up assessment at 2 years of corrected age. The paediatric neurological examination classified the presence or absence of cerebral palsy, according to Palisano et al<sup>25</sup> The time points of the assessments are summarised in Table 2.

## 2.4 | Quantification of intervention sessions

The number and duration of all the study sessions were documented. Two sessions for each infant were video-recorded, to assess any differences between the interventions and to determine what percentage of time was spent on physiotherapy actions and situations. These were recorded 1 and 5 months after the intervention started and the mean scores were used for further analyses, as the contents of the two videos were largely similar.

To classify the intervention elements and to determine their relative quantity, we used the Groningen Observer Protocol, version two<sup>26</sup> and the software programme Observer XT, version 11.5 (Noldus). The Protocol quantifies specific physiotherapy actions as distinguishable intervention elements in terms of their relative duration during a physiotherapy session. Actions are divided into five main categories: neuromotor actions, educational actions towards caregivers, communication, position and situation. Within each category, specific physiotherapeutic intervention elements are defined. The Protocol can also record some independent variables, such as involvement of family members and caregiver roles. Two research assistants, blinded to the group allocation, classified all the videos independently and had good to excellent inter-observer reliability, measured by an interclass correlation coefficient two-way random model (Tables 3 and S1).

**TABLE 1** Background of studied groups

	COPCA (n = 8)	Standard care (n = 8)
Sex (M/F)	6/2	7/1
Gestational age (weeks): median (range)	27 (25-30)	29.5 <sup>a</sup> (26-31)
Birthweight (grams): median (range)	850 (570-1450)	1025 (690-1400)
pH < 7.05	0 (0)	1 (12.5)
Apgar 5 min: median (range)	6.5 (3-8)	7 (3-8)
Firstborn, n (%)	7 (87.5)	3 (37.5)
Twins, n (%)	2 (25)	2 (25)
Bronchopulmonary dysplasia, n (%)	0 (0)	2 (25)
Length of hospital stay (days): mean (SD)	76.4 (30.2)	54.1 (30.4)
Brain lesions		
IVH, grade 1-2 n (%)	1 (12.5)	1 (12.5)
PVL n (%)	1 (12.5)	0
Abnormalities in the neurological examination	8 (100)	8 (100)
General movements at 3-mo CA <sup>b</sup>		
Mildly abnormal, n (%)	5 (62.5)	5 (62.5)
Definitely abnormal, n (%)	0	0
Maternal age (years): median (range)	36.5 (28-49)	30.5 (24-40)
Maternal education, <sup>c</sup> low/middle/high, n (%)	3 (37.5), 1 (12.5), 4 (50.0)	2 (25), 3 (37.5), 3 (37.5)
Corrected age at baseline (weeks): median (range)	4.5 (0-15)	14.5 (0-22)

Note: Significance level: *P* values < .05.

Abbreviations: CA, corrected age; COPCA, Coping with and Caring for Infants with Special Needs; IHV, intraventricular haemorrhage; PVL, periventricular leukomalacia.

<sup>a</sup>Difference between COPCA and standard care: Hodges-Lehmann estimator: +2.0 (95% CI 0.0-5.0).

<sup>b</sup>According to Hadders-Algra et al<sup>28</sup>, this study indicated that not only infants with definitely abnormal GMs at 3-mo CA are at increased risk of developmental disorders, but also infants with the combination of abnormalities at the neurological examination and mildly abnormal GMs.

<sup>c</sup>Levels of education: low = primary education/junior vocational training, middle = secondary education/senior vocational training, high = university education/vocational colleges.

## 2.5 | Data analyses

The power calculation was based on the primary outcome measure, the IMP total score. We calculated that a sample size of 19 infants in both of the groups would have resulted in a power of 80% ( $\alpha = 0.05$ ) to detect a clinically relevant change of 7.5 percentage points in the IMP total score with a standard deviation of 8.2 (10).

Based on the power calculation and anticipating substantial attrition, we had aimed to recruit 68 infants over a 2-year period. However, we were only able to recruit two groups of eight infants, which only allowed us to carry out an RCT with an exploratory analysis. Enrolment had to be stopped before the planned number of infants had been included, as the project's resources ran out due to very slow recruitment.

The characteristics of the groups were quantified by descriptive statistics. The chi-square test was used to compare differences in categorical data at baseline. The chi-square test was used to compare differences in categorical data at baseline. To compare metric data, we computed non-parametric 95% or 99% confidence intervals (CI) for the difference in location parameters, derived from exact Mann-Whitney *U* tests (Hodges-Lehmann estimator). Non-parametric analyses were used for all outcomes, as most data were not normally distributed.

As the primary data analyses indicated that the variation and performance of the normally distributed IMP domains showed interesting results, we fitted a linear mixed model to the data. The model included time, group and the time-group interaction as fixed effects and subject as the random intercept. In the analysis of strongly age-dependent IMP performance scores,<sup>27</sup> we also adjusted for gestational age and corrected age at baseline. Regression analyses were conducted to explore associations between physiotherapy actions and IMP domain scores at 18 months. The confidence level for all estimated effects was adjusted for multiple testing and *P* values of  $\leq .01$  were considered statistically significant.

For the linear mixed model, we used the lme4 package for R and R version 3.6.1 (R Foundation). The other analyses were performed with SPSS, version 26.0 (IBM Corp).

## 3 | RESULTS

### 3.1 | Participants

All 16 infants completed the study up to 18 months of corrected age, but one COPCA group infant was not assessed at 2 years of corrected age (Figure 1). Their characteristics are described in Table 1.<sup>28</sup> All infants had shown neurological abnormalities at inclusion. At 3 months of corrected age, 10 presented with mildly abnormal general movements, but none had definite abnormal general movements. Despite randomisation, gestational age at birth in the COPCA group was significantly lower than in the standard care group (median 27 vs 29.5 weeks) (Table 1). The COPCA group received 15.0-21.0 (mean 18.5) sessions, lasting 23.0-40.5 minutes (mean 37.0 minutes) per session. The standard care group received 11.0-30.0 (mean 19.5) sessions, lasting 28.0-39.5 minutes (mean 32.0 minutes) per session. The overall intensity of therapy did not differ significantly between the groups. None of the infants received other forms of early intervention between baseline and 18 months of corrected age.

All the analyses were performed according to the assigned groups and the results are summarised in Figure 1. The IMP had



**FIGURE 2** Characteristics of the COPCA intervention and standard care infant physiotherapy intervention. A, COPCA intervention. The mother (left) is playing with the infant. She challenges the infant to self-produced motor behaviour while receiving hints from the COPCA coach. B, Standard care. The mother (left) is applying hands-on facilitation techniques to promote the infant's normal sensorimotor experiences while receiving strict instructions from the therapist

Measurements measures	Intervention period			Follow-up PPT	Follow-up DPC
	Baseline	After 3 mo	After 6 mo	At 18 mo of corrected age	At 24 mo of corrected age
Primary outcome					
IMP	+ <sup>a</sup>	+	+	+	
Secondary outcomes					
Family Empowerment Scale	+	+	+	+	
German PEDI			+	+	
Measure of Processes of Care		+	+		
BSID-III					+
Neurological examination					+

**TABLE 2** Primary and secondary outcome measures at the various time points

Abbreviations: BSID-III, Bayley Scales of Infant and Toddler Development, Third Edition; DPC, Developmental Pediatric Center; IMP, Infant Motor Profile; PEDI, Pediatric Evaluation of Disability Inventory, German version; PPT, Pediatric Physical Therapy.

<sup>a</sup>In infants  $\geq 3$  mo of corrected age.

missing data at baseline, as six infants in the COPCA group and three infants in the standard care group were too young to perform it. The Measure of Processes of Care had high levels of missing data and could not be analysed, as the caregivers reported that many items did not apply to their situation.

The neurological examination at 2 years revealed that none of the children had been diagnosed with cerebral palsy.

### 3.2 | Primary and secondary outcomes

The medians and ranges of the primary outcomes in both groups at all time points are summarised in Tables 4 and S2 also includes the secondary outcomes. The IMP total scores and adaptability, symmetry and fluency domain scores did not differ between the groups. The IMP scores for the variation and performance domains were similar in the two groups at baseline and 3 and 6 months later, but higher in the COPCA group than the standard group at 18 months

of corrected age. They were a median of 95.0 vs 88.5, with a Hodges-Lehmann estimator of 7.0 (95% CI 0.0-13.0), for variation. The performance values were 93.5 vs 91.5 and 2.0 (95% CI 1.0-4.0), respectively.

The total Family Empowerment Scale scores did not differ significantly between the groups. The German PEDI scores for both groups did not differ at baseline and 3 months later. However, 6 months after baseline the median functional mobility score of the standard care group was significantly higher than the COPCA group (43.9 vs 28.6), with a Hodges-Lehmann estimator of 9.0 (95% CI 3.6-30.5). At 18 months of corrected age, the difference had disappeared. The BSID-III scores at 2 years of corrected age were not significantly different between the groups.

We carried out linear mixed model analyses of the IMP variation and performance domains (Table 5). These indicated a significant improvement in the mean variation domain score for the COPCA group between 3 and 6 months after baseline, by 14.2 (95% CI 6.1-22.3). This was nine percentage points (95% CI 0.3-17.5) more than

**TABLE 3** Percentages of time spent on physiotherapy actions and situations for COPCA intervention and standard care

PPT actions and situations	ICCs (n = 16) <sup>a</sup>	COPCA (n = 8) Median % (range)	Standard care (n = 8) Median % (range)
<b>Neuromotor actions</b>			
Facilitation techniques	0.89	5.2 (1.0-7.9)	<b>15.4 (4.9-23.9)</b>
SPMB	0.95	<b>40.6 (34.9-66.6)</b>	22.2 (16.5-41.8)
Involved in exploration	0.86	18.3 (10.4-38.3)	15.8 (4.7-26.5)
No signs of exploration	0.93	<b>25.6 (20.1-28.4)</b>	11.7 (0.5-19.3)
CSPMB, infant is allowed to continue activity	0.93	<b>37.2 (9.5-41.5)</b>	10.4 (3.5-21.7)
CSPMB, flows over into hands-on techniques	0.92	6.3 (0.0-14.9)	<b>27.3 (11.5-56.9)</b>
<b>Educational actions towards caregiver</b>			
Caregiver coaching	0.87	<b>90.0 (53.5-99.2.0)</b>	0.0 (0.0-0.0)
Caregiver training	0.80	0.0 (0.0-1.4)	<b>51.5 (0.0-68.4)</b>
Not specified educational actions <sup>b</sup>	0.75	10.0 (0.8-46.5)	<b>48.5 (31.6-100.0)</b>
<b>Communication</b>			
Instruct	0.80	6.6 (4.6-16.9)	5.2 (0.9-11.3)
Strict instruction	0.76	3.1 (1.0-11.9)	3.4 (0.0-7.8)
Giving hints	0.78	<b>4.5 (1.8-5.8)</b>	2.1 (0.3-3.5)

Note: % = Median percentage of time spent on physical therapy actions and situations during physiotherapy sessions 1 and 5 mo after the start of intervention; differences between groups tested with Mann-Whitney U tests (non-parametric confidence intervals for the difference in location parameters, ie Hodges-Lehmann estimator).

Actions preceded by cubed numeration are modifiers (specifications of the associated physiotherapeutic intervention element).

$P < .01$ —bold number shows statistically significant  $P$  values.

Abbreviations: COPCA, Coping with and Caring for Infants with Special Needs; CSPMB, challenged to self-produce motor behaviour; SPMB: self-produce motor behaviour.

<sup>a</sup>ICC: interclass correlation coefficient; ICCs were calculated to assess inter-rater reliability; they are based on the scores of two independent research assistants who assessed all videotapes.

<sup>b</sup>'Not specified educational actions' were scored if neither training nor coaching were taking place. Examples of this behaviour are the situation during which the therapist is treating the infant without information exchange with the caregiver or the situation that the therapist is occupied with something not related to the session.

the standard care group (Figure 3). The performance domain scores for both groups improved between 3 and 6 months after baseline and between 3 months after baseline and 18 months of corrected age (Table 5). This time-group interaction indicated that the COPCA group improved by 12 percentage points (95% CI 4.1-20.6) more between 3 months after baseline and 18 months of corrected age than the standard care group (Figure 3).

### 3.3 | Session contents and association with the IMP

The percentage of time spent on neuromotor actions, educating caregivers and communication during the video-recorded sessions are summarised in Table 3, with all physiotherapy actions covered in Table S1. In the COPCA group, most of treatment time spent on neuromotor actions focused on self-produced motor behaviour (median 40.6%) and hands-off techniques that challenged the infants to

self-produce motor behaviour (median 37.2%). Most of the time in the educating caregiver category was spent on coaching caregivers (median 90.0%).

In the standard care group, most of treatment time on neuromotor actions was spent on challenging the infant to self-produce motor behaviour flowing over into hands-on technique within 20 seconds (median 27.3%). Facilitation, another hands-on technique, was also frequently applied (median 15.4%). Most of the educating caregiver category was spent on training them (median 51.5%), with no time spent on coaching. The time spent on neuromotor and educational actions differed significantly between the groups (Table 3).

In the 16 video-recorded COPCA sessions, 15 caregivers played an active role and engaged with the infant in daily routines like playing, while the therapist observed the interaction and provided hints and suggestions. One caregiver played an observational role in the treatment while she simultaneously cared for the twin brother. In the 16 video-recorded standard care sessions, eight caregivers

TABLE 4 IMP scores in both intervention groups at all measurement moments

	Baseline		After 3 mo		After 6 mo		At 18 mo of corrected age		At 24 mo of corrected age				
	COPCA Median (range)	Standard care Median (range)	COPCA Median (range)	Standard care Median (range)	COPCA Median (range)	Standard care Median (range)	COPCA Median (range)	Standard care Median (range)	COPCA Median (range)	Standard care Median (range)			
IMP total score	n = 2 74.5 (71-78)	n = 5 80 (69-83)	n = 8 77.5 (67-81)	n = 8 79 (73-86)	n = 8 86 (80-91)	n = 8 84 (79-94)	n = 8 92.5 (85-98)	n = 8 90.5 (85-98)	n = 8 95 (87-100)	n = 8 88.5 (77-96)	n = 8 90.5 (85-98)	n = 8 90.5 (85-98)	n = 8 90.5 (85-98)
IMP variation	n = 2 83.5 (79-88)	n = 5 88 (77-97)	n = 8 81 (66-94)	n = 8 84 (64-91)	n = 8 85 (75-100)	n = 8 90.5 (77-100)	n = 8 95 (87-100)	n = 8 88.5 (77-96)	n = 8 95 (87-100)	n = 8 88.5 (77-96)	n = 8 90.5 (85-98)	n = 8 90.5 (85-98)	n = 8 90.5 (85-98)
IMP adaptability	n = 0 NA	n = 0 NA	n = 2 80.5 (75-86)	n = 5 75 (75-86)	n = 8 87 (79-93)	n = 7 81 (70-94)	n = 8 92 (79-96)	n = 8 90 (83-100)	n = 8 92 (79-96)	n = 8 90 (83-100)	n = 8 90 (83-100)	n = 8 90 (83-100)	n = 8 90 (83-100)
IMP symmetry	n = 2 90.5 (87-94)	n = 5 95 (71-100)	n = 8 90 (81-100)	n = 8 97 (71-100)	n = 8 100 (92-100)	n = 8 100 (72-100)	n = 8 100 (88-100)	n = 8 98 (92-100)	n = 8 100 (88-100)	n = 8 98 (92-100)	n = 8 98 (92-100)	n = 8 98 (92-100)	n = 8 98 (92-100)
IMP fluency	n = 2 72.5 (70-75)	n = 5 75 (75-83)	n = 8 75 (75-75)	n = 8 75 (75-88)	n = 8 85.5 (75-100)	n = 8 75 (75-100)	n = 8 85 (70-100)	n = 8 90 (70-100)	n = 8 85 (70-100)	n = 8 90 (70-100)	n = 8 90 (70-100)	n = 8 90 (70-100)	n = 8 90 (70-100)
IMP performance	n = 2 51 (49-53)	n = 5 52 (41-64)	n = 8 57.5 (45-67)	n = 8 67.5 (55-86)	n = 8 70.5 (66-88)	n = 8 84 (69-90)	n = 8 93.5 (92-96)	n = 8 91.5 (90-96)	n = 8 93.5 (92-96)	n = 8 91.5 (90-96)	n = 8 91.5 (90-96)	n = 8 91.5 (90-96)	n = 8 91.5 (90-96)

Note: Differences between the groups were tested with the non-parametric confidence intervals for the difference in location parameters (Hodges-Lehmann estimator)—derived from exact Mann-Whitney *U* tests. The values of this evaluation are not listed in this table.

*P* < .05—bold number shows statistically significant *P* values.

Abbreviations: COPCA, Coping with and Caring for Infants with Special Needs; IMP, Infant Motor Profile; MAC, median corrected age in month; NA, not assessed.



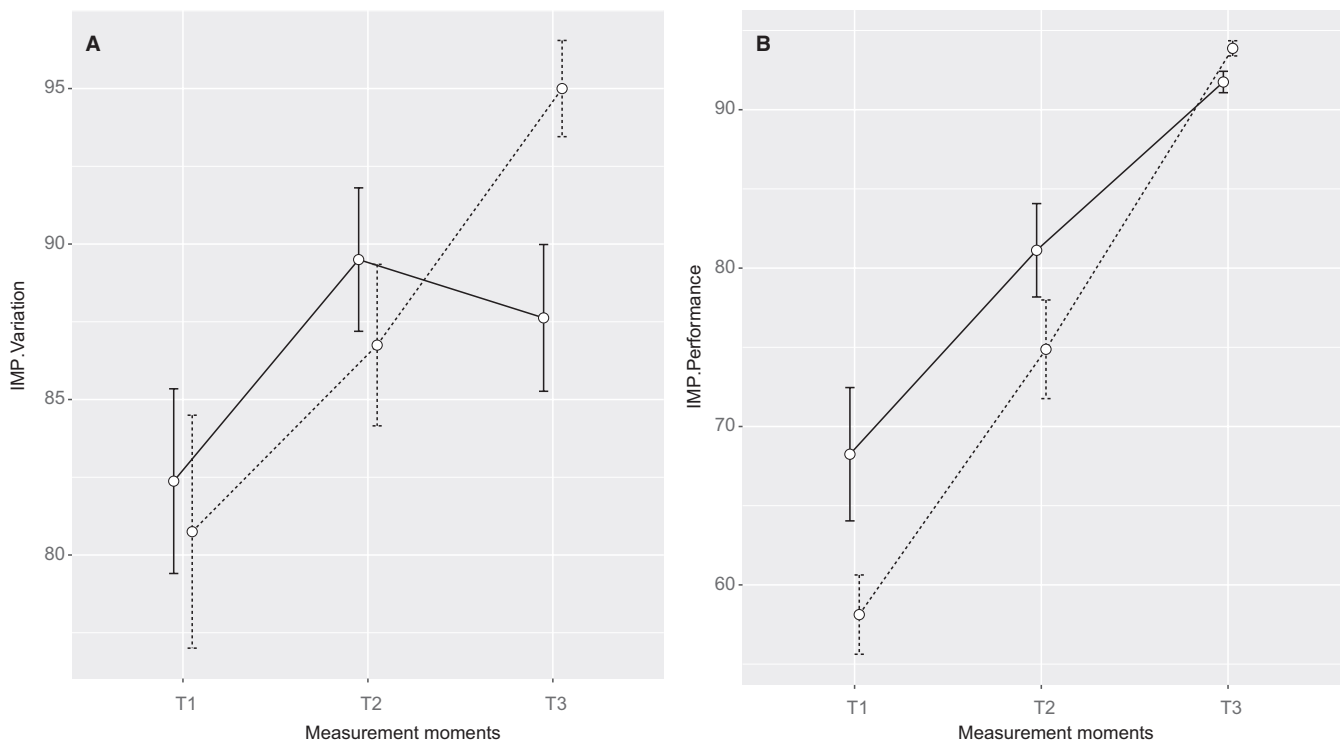
**TABLE 5** Within-group time contrasts estimated from a linear mixed model

	Between baseline and after 3 mo		Between 3 and 6 mo after baseline		Between 3 mo after baseline and 18 mo of corrected age	
	COPCA	Standard care	COPCA Mean (95% CI)	Standard care Mean (95% CI)	COPCA Mean (95% CI)	Standard care Mean (95% CI)
IMP variation	NA	NA	n = 8 6.0 (-2.1-14.1)	n = 8 7.1 (-1.0-15.2)	n = 8 <b>14.2 (6.1-22.3)</b>	n = 8 5.2 (-2.8-13.3)
IMP performance	NA	NA	n = 8 <b>16.8 (9.0-24.5)</b>	n = 8 <b>12.9 (5.1-20.6)</b>	n = 8 <b>35.8 (28.0-43.5)</b>	n = 8 <b>23.5 (15.8-31.2)</b>

Note: Performance adjusted for gestational age and corrected age at baseline, confidence level adjusted for multiple testing (Tukey's method).

$P < .01$ —bold number shows statistically significant changes.

Abbreviations: CI, confidence interval; COPCA, Coping with and Caring for Infants with Special Needs; NA, not assessed.



**FIGURE 3** Developmental changes in IMP scores in of both groups. (A) developmental changes in the variation domain. (B) developmental changes in the performance domain. The lines indicate the group mean values, and the error bars indicate one standard error. Group: -----, COPCA; ———, Standard care

played an observational role, six guided the attention of the infants while the therapist performed the treatment and two performed the handling techniques by following the therapist's instructions.

We only present the exploratory regression analyses of associations between physiotherapeutic actions and infants in the IMP variation and performance domains at 18 months of corrected age. This is because these were the only outcome variables to show differences in the between-group analyses. The IMP variation score was negatively associated with the neuromotor action of challenging the infant to self-produced motor behaviour, flowing over into hands-on techniques (beta  $-0.27$ , 95% CI  $-0.45$  to  $-0.09$ ,  $P = .005$ ) and the educational action of caregiver training (beta  $-0.14$ , 95% CI  $-0.22$  to  $-0.03$ ,  $P = .01$ ). However, the educational action of caregiver

coaching (beta  $0.03$ , 95% CI  $0.01$ - $0.05$ ,  $P = .01$ ) showed a positive association with IMP performance scores.

## 4 | DISCUSSION

This small, explorative RCT showed that preterm infants born before 32 weeks of gestation without significant brain lesions showed some better motor outcomes at 18 months of corrected age if they received 6 months of COPCA rather than standard care. These improvements were in the IMP domains of variation and performance. COPCA and standard care differed significantly with regard to neuromotor actions and caregiver education actions. Hands-on

techniques and caregiver training were negatively associated with the IMP variation domain and caregiver coaching was positively associated with the IMP performance domain. The latter association was not very strong, but confirms previous COPCA studies,<sup>6,9,11</sup> which highlights the significance of this intervention element in clinical practice.

The finding that COPCA was associated with better IMP scores in the variation and performance domains at 18 months agrees with Sgandurra et al<sup>18</sup> on the effect of early interventions in very preterm infants without brain lesions. Our linear mixed model analyses indicated, that the variation domain scores, which reflect the size of the motor repertoire, continuously increased between baseline and the follow-up, at 18 months of corrected age, in the COPCA group. This was particularly true between the end of the intervention and this follow-up. In contrast, the motor repertoire of the infants in the standard care group increased between baseline and the end of the intervention, but had decreased again by follow-up at 18 months of corrected age. These results may be interpreted as follows. First, early interventions may increase the motor repertoire of preterm infants at moderate-to-high risk of developmental disorders. Second, the effects are sustainable because caregivers who appreciated the importance of challenging their infant's self-produced motor behaviour continue to act accordingly, even after the end of the intervention. According to the neuronal group selection theory,<sup>7</sup> variation is a result of explorative activity of the nervous system. This means that the size of an infant's motor repertoire is associated with their level of experience and environmental influences. This idea is supported by our finding that challenging the infant to self-produce motor behaviour, flowing over into hands-on techniques, was associated with lower scores in the IMP variation domain. This is because the hands-on technique interferes with the infant's own exploratory efforts and thereby reduces their motor experiences. However, seriously reduced IMP variation scores have been associated with structural brain anomalies.<sup>7</sup> Fortunately, most moderate- to high-risk preterm infants do not have severe brain lesions.<sup>1</sup> Nevertheless, birth before 32 weeks has been associated with immaturity and increased stress,<sup>29</sup> which may interfere with explorative activity. This study and the study by Sgandurra et al<sup>18</sup> indicate that if preterm infants have a moderate-to-high risk, it is possible to increase their motor repertoire through enhanced explorative activity. This is achieved by challenging their self-produced motor behaviour and providing an enriched environment.

Our study showed that COPCA had a beneficial effect on both the IMP variation and performance domain scores. The performance score summarises the achievement of developmental motor milestones, which is an important aspect for daily living activities and a marker for the sustainable effect of the COPCA intervention.

The process evaluation supported our suggestion that the relatively late positive effect of the COPCA intervention programme was mediated by the family. This analysis revealed that, at 18 months of corrected age, parent training was associated with worse motor outcomes, while parent coaching was associated with better motor outcomes. Caregiver training and coaching are two different

approaches with different goals, beliefs and attitudes.<sup>30</sup> In this study, they also produced different motor outcomes in the infants. Our results were in line with Blauw-Hospers et al,<sup>6</sup> who found that COPCA's caregiver coaching was associated with better functional mobility at 18 months of corrected age. Caregiver coaching appears to be sustainable, as it empowers caregivers to support the infant's long-term development after the end of the intervention.

The higher German PEDI functional mobility score in the standard care group at the end of the intervention was most likely due to the higher corrected age of the infants in this group: a mean age of 10 vs 7 months in the COPCA group. The mean social function score of both groups was one to two standard deviations below the average mean of a normative sample. This echoes other studies that reported that preterm infants born before 32 weeks were at risk of impaired social behaviour.<sup>2</sup> The cognitive and the motor composite score of the BSID-III at 24 months of corrected age did not differ between the groups and was within one standard deviation of the normative mean in both groups. However, assessing cognitive and motor outcome around the age of 2 years should be interpreted with caution, as their predictive value for motor and cognitive outcomes at school age is limited. Cognitive and motor delays may become obvious later in life, when demands for cognition and motor skills increase and subtle impairments can become apparent. We could not confirm our hypothesis, that the COPCA intervention would show positive effects on family outcome and offer two explanations. The first relates to the Family Empowerment Scale, which primarily measures empowerment as a current state and is not related to an early intervention process. It was originally developed for the families of children with emotional disabilities, which is quite a different context to our study. This may imply that the questions it posed do not measure empowerment in the families of young infants who need early interventions. The second relates to the Measure of Processes of Care, which was designed to measure how families and children with diagnosed disabilities experience health care during multidisciplinary rehabilitation treatment centre. This implies that the intended context differed from the one in our exploratory RCT. Our participants were the caregivers of extremely and very preterm infants at moderate-to-high risk of developmental disorders, without a developmental diagnosis at the beginning of the intervention process. In such a situation, caregivers usually sail between hope and anxiety. All families received individual face-to-face interventions and no other therapies. This means that they did not receive services from a multidisciplinary rehabilitation treatment centre and that they did not respond to the questions related to a rehabilitation centre context or a manifest disability. Despite this, we chose both instruments as they were the best available.

The strengths of this study were the double approach of the RCT and process evaluation and the longitudinal design. The process evaluation allowed for detailed quantitative content analyses of the two intervention methods and established associations between intervention elements and outcomes. The longitudinal design meant that we could use a linear mixed model analysis to show the sustainability of coaching.

The study had several limitations. The small sample sizes meant the study was underpowered and enrolment had to be stopped before the planned number of infants had been included, as the project's resources ran out due to very slow recruitment. We learnt that recruiting for this type of early intervention RCT depends on interfering trials and on communication with caregivers and between multiple health professionals with different roles and high workloads. Despite randomisation, gestational age at birth was higher in the standard care than COPCA group. This might have been an advantage for the former, but we did adjust for gestational age and corrected age in the linear mixed model analyses. An additional limitation was the missing data for the primary outcome of IMP at baseline. Some infants needed the intervention before 3 months of corrected age, but were too young to perform the IMP at baseline. It would have been better to use an instrument such as the Alberta Infant Motor Scale, which covers term age to 18 months. However, the major drawback of this scale is that it has less sensitivity than the IMP to measure the effects of early interventions.<sup>18</sup> Finally, the last assessment was at 2 years of age and this might have been a limitation, as it is well known that after the age of 2 years children still may grow into, or out of, deficits.

## 5 | CONCLUSION

This explorative study of preterm infants born before 32 weeks without significant brain lesions showed that the COPCA intervention programme was associated with a better motor outcome at 18 months of corrected age than standard care. The process evaluation indicated that the key elements that contributed to this differential outcome were caregiver coaching and not interfering in the child's motor activities with hands-on techniques. However, future studies with adequate power and long-term follow-up are needed to validate and confirm the promising results of this study.

A number of instruments need to be developed in the near future to measure a number of factors. These are reliable, applicable and sensitive family-related outcomes, perceptions of family centring in families with infants and very young children, family engagement and well-being, parents' confidence, competence and capacity.

### ACKNOWLEDGEMENT

The authors would like to thank all the therapists, infants and parents who participated in the study.

### CONFLICT OF INTEREST

The authors have no conflicts of interest to declare.

### ETHICAL APPROVAL

The study was approved by the ethic committees in Zurich, east Switzerland and northwest and central Switzerland. The trial was registered in the German register for clinical trials and in the World Health Organisation International Clinical Trials Registry Platform (DRKS00009794).

### ORCID

Schirin Akhbari Ziegler  <https://orcid.org/0000-0001-8640-0774>  
Mijna Hadders-Algra  <https://orcid.org/0000-0001-6845-5114>

### REFERENCES

- Pascal A, Govaert P, Oostra A, Naulaers G, Ortibus E, Van den Broeck C. Neurodevelopmental outcome in very preterm and very-low-birthweight infants born over the past decade: a meta-analytic review. *Dev Med Child Neurol*. 2018;60(4):342-355.
- de Kieviet JF, Piek JP, Aarnoudse-Moens CS, Oosterlaan J. Motor development in very preterm and very low-birth-weight children from birth to adolescence: a meta-analysis. *JAMA*. 2009;302(20):2235-2242.
- Data Collection. Swiss Society of Neonatology [Internet]. <https://www.neonet.ch/swissneonet/data-collection>. Accessed May 3, 2020
- Spittle A, Orton J, Anderson PJ, Boyd R, Doyle LW. Early developmental intervention programmes provided post hospital discharge to prevent motor and cognitive impairment in preterm infants. *Cochrane Database Syst Rev*. 2015;(11):CD005495.
- Dunst CJ, Trivette CM, Hamby DW. Meta-analysis of family-centered helping practices research. *Ment Retard Dev Disabil Res Rev*. 2007;13(4):370-378.
- Blaauw-Hospers CH, Dirks T, Hulshof LJ, Bos AF, Hadders-Algra M. Pediatric physical therapy in infancy: from nightmare to dream? A two-arm randomized trial. *Phys Ther*. 2011;91(9):1323-1338.
- Hadders-Algra M. Early human motor development: from variation to the ability to vary and adapt. *Neurosci Biobehav Rev*. 2018;90:411-427.
- Hielkema T, Hamer EG, Reinders-Messelink HA, et al. LEARN 2 MOVE 0-2 years: effects of a new intervention program in infants at very high risk for cerebral palsy; a randomized controlled trial. *BMC Pediatr*. 2010;10:76.
- Hielkema T, Blaauw-Hospers CH, Dirks T, Drijver-Messelink M, Bos AF, Hadders-Algra M. Does physiotherapeutic intervention affect motor outcome in high-risk infants? An approach combining a randomized controlled trial and process evaluation. *Dev Med Child Neurol*. 2011;53(3):e8-e15.
- Hielkema T, Hamer EG, Boxum AG, et al. LEARN2MOVE 0-2 years, a randomized early intervention trial for infants at very high risk of cerebral palsy: neuromotor, cognitive, and behavioral outcome. *Disabil Rehabil*. 2019;1-10. <https://doi.org/10.1080/09638288.2019.1610508> [publishes online ahead of print, 2019 May 13].
- Hielkema T, Boxum AG, Hamer EG, et al. LEARN2MOVE 0-2 years, a randomized early intervention trial for infants at very high risk of cerebral palsy: family outcome and infant's functional outcome. *Disabil Rehabil*. 2019;1-9. <https://doi.org/10.1080/09638288.2019.1610509> [publishes online ahead of print, 2019 May 29]
- Akhbari Ziegler S, Mitteregger E, Hadders-Algra M. Caregivers' experiences with the new family-centred paediatric physiotherapy programme COPCA: a qualitative study. *Child Care Health Dev*. 2019;46(1):28-36.
- Akhbari Ziegler S, Dirks T, Hadders-Algra M. Coaching in early physical therapy intervention: the COPCA program as an example of translation of theory into practice. *Disabil Rehabil*. 2019;41(15):1846-1854.
- Hoffmann TC, Glasziou PP, Boutron I, et al. Better reporting of interventions: template for intervention description and replication (TIDieR) checklist and guide. *BMJ*. 2014;348:g1687.
- Akhbari Ziegler S, Dirks T, Reinders-Messelink HA, Meichtry A, Hadders-Algra M. Changes in therapist actions during a novel paediatric physical therapy program: successes and challenges. *Pediatr Phys Ther*. 2018;30(3):223-230.

16. Heineman KR, Bos AF, Hadders-Algra M. The infant motor profile: a standardized and qualitative method to assess motor behaviour in infancy. *Dev Med Child Neurol.* 2008;50(4):275-282.
17. Heineman KR, Middelburg KJ, Bos AF, et al. Reliability and concurrent validity of the infant motor profile. *Dev Med Child Neurol.* 2013;55(6):539-545.
18. Sgandurra G, Lorentzen J, Inguaggiato E, et al. A randomized clinical trial in preterm infants on the effects of a home-based early intervention with the "CareToy System". *PLoS One.* 2017;12(3):e0173521.
19. Koren PE, DeChillo N, Friesen BJ. Measuring empowerment in families whose children have emotional disabilities: a brief questionnaire. *Rehabil Psychol.* 1992;37(4):305-321.
20. Haley SM, Coster WJ, Ludlow LH, Haliwanger J, Andrellos P. *Pediatric Evaluation of Disability Inventory: Development, Standardization, and Administration Manual.* Boston: New England Medical Center Hospital/ Trustees of Boston University; 1992.
21. Haley SM, Coster WJ, Ludlow LH, Haliwanger J, Andrellos P. PEDI-D Pediatric Evaluation of Disability Inventory. Assessment zur Erfassung von Aktivitäten des täglichen Lebens bei Kindern mit und ohne Beeinträchtigung. (C. Schulze, J. Page, Trans. and Eds.). Idstein: Schulz-Kirchner; 2014 (Edition VITA ACTIVA).
22. King S, Rosenbaum P, King G. *The Measure of Processes of Care: A Means to Assess Family-centred Behaviours of Health Care Providers.* Hamilton, ON: McMaster University, Neurodevelopmental Clinical Research Unit; 1995.
23. Iten N, Kraus de Camargo O. The Measure of Processes of Care (MPOC-56 (R) D.04). © 1995 S. King, Rosenbaum and G. King. CanChild for Childhood Disability Research, McMaster University.
24. Bayley N. *Bayley Scales of Infant and Toddler Development*, 3rd edn. San Antonio, TX: Harcourt Assessment; 2006.
25. Palisano RJ, Hanna SE, Rosenbaum PL, et al. Validation of a model of gross motor function for children with cerebral palsy. *Phys Ther.* 2000;80(19):974-985.
26. Sonderer P, Akhbari Ziegler S, Gressbach Oertle B, Meichtry A, Hadders-Algra M. Psychometric properties of a standardized observation protocol to quantify pediatric physical therapy actions. *Pediatric Phys Ther.* 2017;29(3):244-250.
27. Heineman KR, Bos AF, Hadders-Algra M. Infant Motor Profile and cerebral palsy: promising associations. *Dev Med Child Neurol.* 2011;53(Suppl 4):40-45.
28. Hadders-Algra M, Mavinkurve-Groothuis AM, Groen SE, Stremmelaar EF, Martijn A, Butcher PR. Quality of general movements and the development of minor neurological dysfunction at toddler and school age. *Clin Rehabil.* 2004;18(3):287-299.
29. Weber A, Harrison TM. Reducing toxic stress in the neonatal intensive care unit to improve infant outcomes. *Nurs Outlook.* 2019;67(2):169-189.
30. Akhbari Ziegler S, Hadders-Algra M. Coaching approaches in early intervention and paediatric rehabilitation. *Dev Med Child Neurol.* 2020;62(5):569-574.

### SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section.

**How to cite this article:** Akhbari Ziegler S, von Rhein M, Meichtry A, Wirz M, Hielkema T, Hadders-Algra M; the Swiss Neonatal Network & Follow-Up Group. The Coping with and Caring for Infants with Special Needs intervention was associated with improved motor development in preterm infants. *Acta Paediatr.* 2020;00:1-12. <https://doi.org/10.1111/apa.15619>