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A Physical Therapy Intervention to Advance Cognitive and Motor Skills: A Single Subject Study of a Young Child with Cerebral Palsy

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Introduction

Early intervention (EI), the United States' educational intervention provided to children birth to 3 years old with delays, is designed to reduce the severity of developmental impairments and enhance a child's ability to fully participate in family and society.[1] However, evidence that EI affects developmental or educational outcomes for children with severe neuromotor dysfunction is limited. A systematic review on early motor intervention for infants at high risk for cerebral palsy found limited evidence of intervention effectiveness. [2] However, a review found promising evidence for effective motor interventions that incorporated child-initiated movement, parent education, and environmental modification. Interventions designed to advance cognitive and motor skills in children with severe motor impairment ware less common than those targeting motor skills alone. [3,4]

Infants who experience neonatal encephalopathy are at high risk for global developmental impairments. While the rate of motor and cognitive impairments has decreased since the introduction of neonatal hypothermia, half of infants with neonatal encephalopathy will go on to have an IQ less than 85 and 27% will have an IQ less than 70. However, 96% of infants with cerebral palsy (CP) following neonatal encephalopathy have an IQ less than 70.[5] While the rate of cognitive impairment is likely very high in children post neonatal encephalopathy, valid assessment of cognition is difficult in young children with severe motor impairments.[6] The majority of outcome measures for children less than 4 years of age require manipulation of objects such as uncovering hidden objects, placing objects in and out of containers, or completing puzzles.[7] For a child with limited postural control or hand function, these tasks are challenging, making it unclear if the score reflects cognitive or motor impairment. Currently there is no standard for testing cognitive abilities in young children with CP.[6] Serial assessment is recommended to quantify change over time. Assessing a combination of motor and cognitive outcomes following intervention will lead to greater understanding of the global benefits of therapy services.

While pediatric physical therapy was born from neuromaturational perspectives, modern theories support the interconnection between body systems and developmental domains.[8] Dynamic systems theory supports the critical, but equal, importance of body systems including the central nervous, musculoskeletal, and cardiorespiratory systems in development. [9] Perception Action Theory[10] and Grounded Cognition[11] support the inter-relationship between movement and learning. All 3 theories support the importance of the task, environment, and individual's characteristics in supporting/impeding a child's development. There continues to be limited evaluation of the impact of physical therapy using a perception action or grounded cognition approach to advance motor and cognitive outcomes.[12,13] While many EI program are moving toward a transdisciplinary intervention approach, there is little training in Doctor of Physical Therapy programs on the relationship between motor and cognitive skills to prepare physical therapist for this role in practice.

In addition to the need for measuring cognitive outcomes, these theoretical models all suggest the importance of engaging parents in intervention to allow for daily practice and environmental modification to support learning. [12,13] However, few studies measure changes

in parent child interaction or environmental modification, which may influence the outcomes of intervention. [14,15]

The Sitting Together And Reaching To Play (START-Play) intervention is designed to incorporate child-initiated movement, parent education, and environmental modification to enhance both cognitive and motor exploration.[16] Sitting and reaching are used to scaffold opportunities to explore toys, problem-solve, and gain independence with initiating interactions. This intervention program was designed for infants who demonstrate some ability to prop sit or who can sit independently, but cannot get in and out of sitting.[16] Given the dearth of evidence-based interventions to jointly advance motor and cognitive skills in children with severe motor impairments, this study was designed to evaluate the potential of the START-Play intervention to improve motor and cognitive outcomes in a child with severe motor impairments. Thus, the purpose of this study was to evaluate the change in motor and cognitive abilities of a single child during participation in the START-Play intervention.

Methods

Subject:

The child, hereafter referred to as “C”, was born at term and diagnosed with hypoxic ischemic encephalopathy requiring resuscitation, full body hypothermia and additional medical interventions during a 36 day stay in the neonatal intensive care unit (NICU). Physical therapy in the NICU focused on parent support and encouraging active movement. C had poor oral motor skills and aspiration during oral feedings requiring a gastrostomy tube. C was followed by the Neonatal Continuing Care Program from NICU discharge until 2 years of age. She was referred to the CP clinic and pediatric orthopedics for ongoing monitoring. C’s mother reported ongoing medical management of her gastrointestinal pain and seizures during the first year of life. C presented with variable central muscle tone, and increased muscle tone in all extremities. At 12 months of age she was given a diagnosis of probable CP that was confirmed at 24 months of age. Based on her motor function at the end of the study her clinical presentation is that of a child with spastic bilateral cerebral palsy with a Gross Motor Function Classification System level of IV. She has 2 very supportive parents and an older sibling.

C was referred to her local early intervention program at the time of NICU discharge. Her mother and the early intervention team worked together to draft her service plan which included early intervention occupational therapy services 1 time/week and service coordination. These services and the therapist who provided the services were consistent for the first 2 years of C’s life.

Study Design and Outcome Assessment:

An AB phase design without reversal[17] was used to evaluate the efficacy potential of the START-Play intervention to advance cognitive and motor skills in a child with severely impaired motor skills. (Table 1 includes a detailed timeline)

Phase A: Between the age of 4-16 months, C participated in another clinical trial as part of the comparison group providing a baseline for this study as all outcomes were completed by reliable and blinded assessors.[18] C was evaluated for her participation in a second clinical trial at 16 months. Her family consented to participation in both studies. Due to the severity of her motor impairments her data was not included in the analysis for the larger clinical trial. However, she was an ideal participant for a single subject study designed to evaluate the use of START-Play to advance cognitive skills in children with severe motor impairments.

Phase B: C participated in the START-Play intervention for 3 months from 17-20 months of age. With 1 assessment, including some of the outcome measures, completed in the middle of the intervention period.

Follow up: Two follow up visits were completed after the end of the START-Play intervention visits at 23 and 29 months of age . However, the mother reported continuing some activities she learned on her own after the intervention, so a full reversal to the baseline was not possible.

Outcome Measures: The Bayley Scales of Infant and Toddler Development, Third Edition, (Bayley-III) was selected as a primary outcome measure.[7] Change in Bayley-III raw scores was evaluated with at least 2 data points per phase. The raw scores on the Bayley-III were selected for analysis, as they best represent the child's changing abilities. All assessments included in this study were videotaped and scored by blinded research personnel who had achieved intra- and inter-rater agreement of greater than 85% for each measure. [16] Inter and Intra-rater reliability on the Bayley (ICC2,1) was greater than 0.95 for all measures.

Multiple secondary measures were included during the data collections to provide a more detailed picture of C's changing motor and cognitive skills. There were 4 measures completed including baseline, 3 in the intervention, and 3 in the follow up period for each of the secondary measures.

A modified version of the Early Problem Solving Indicator (EPSI), was used to assess the child's self-initiated use of early problem solving skills during a standardized play session. [19,20] C was videotaped interacting with the same 3 toys at each visit, each for 2 minutes while in a sitting position, with as much support as needed to remain sitting. The frequency of problem-solving behaviors was determined using behavioral coding software (Datavyu) and the video of the assessment. Coders quantified the number of looks (gazes at the toy), explores (manipulates the toy), functions (engages the toy functions), and solutions (completes all possible functions and solves the toy). While it is always difficult to identify the best cognitive outcome assessments for young children, previous experience with the EPSI scored using coding software suggest it is sensitive to changes overtime if repeated measures were used to quantify a general progression.

The Gross Motor Function Measure (GMFM) item set version was used to assess global change in motor abilities, while the GMFM-88 sitting subscale measured sitting only.[21] Reaching was measured with the child seated in an infant seat with toys presented at midline

chest height. The percent time with either hand in contact with the toy was recorded as a measure of reaching ability.[22] A blinded assessor scores each GMFM from video with inter and intra-rater reliability ICC(2,1) ranging from 0.91 to 1.00.

The START-Play intervention challenges parents to be active participants in the intervention sessions and to practice the intervention's key concepts between visits.[16] As such, we wanted to include a measure of parent play in order to quantify changes in parent-infant play interactions over time. A 5-minute free play session of the mother and child was video recorded at each assessment session. The mother was asked to play with C however she usually would. Custom behavioral coding was used to assess the frequency of interactions in which the mother presented cognitive opportunities. Cognitive opportunities were defined as opportunities that required the child to work on a cognitive skill such as object affordances, mean end, or object permanence while C was also engaged in a motor action. For example, providing an opportunity to manually explore a toy while in supported sitting on the floor affords discovery of the object properties while also practicing the challenging task of head and trunk control and occasional single arm support. In contrast, shaking a toy while holding a child would not be considered a cognitive opportunity as the child could not engage actively with the toy and the motor task was not a challenge. The behavioral coder maintained a high degree of reliability with greater than 90 percent agreement for this behavior.

Intervention:

The START-Play intervention included twice weekly home visits by a physical therapist for 3 months for a total of 24 one-hour sessions. The interventionist was trained by the START-Play research team and participated in the larger clinical trial, meeting high fidelity standards on adherence to the key principles of the START-Play intervention.[16]

During the START-Play intervention, the therapist and family worked together to provide intensive, individualized, daily activities to advance reaching and sitting. The START-Play intervention utilizes a perceptual-motor framework centered on early cognitive constructs. Intervention occurs in infants' natural environment, using caregiver social support to scaffold infant skills. The key ingredients of the START-Play intervention are: 1) cognitive constructs blended with motor challenges, 2) opportunities for 4 key cognitive constructs (object permanence, means-end understanding, body/object affordances, joint attention) blended with social support, and 3) parents brainstorming and assisting directly with the "just right" challenge of blended motor/cognitive skills. Therapists aim to engage infants and parents in play and problem-solving utilizing variable sitting and reaching abilities while learning about the 4 key cognitive constructs. The specific intervention activities match the skill level of each child from early to more mature skill.

Working with the interventionist, C's mother was guided to discover and problem-solve motor/cognitive challenges as a unit, and to link small motor changes to cognitive advancement for overall developmental progression. [16] Each session began with an update on the activities that C and her mother had worked on since the previous session. The interventionist and C's mother would engage in play activities based on C's interest. Some examples of activities include tracking toys in a variety of positions (early object

permanence), setting up motor tasks to help C be successful at contacting and manually exploring objects (object affordances), and encouraging joint engagement with a toy (precursor to joint attention). Adequate time was provided to allow C to perceive the challenge, organize her motor patterns, and attempt the action. If she was unable to complete the cognitive task the motor challenge was reduced to allow more attention to be focused on the cognitive skill. Other times the cognitive task, such as searching for a missing object, would be easier to allow her to keep working on the cognitive skills and motor skills together. This just- right challenge allowed C and her mother to find success many times in a single session, while balancing motor and cognitive challenges. The mother was encouraged to use the principle of START-Play in her regular play with C, however no specific time or goal for frequency or duration was provided for activities between sessions. Positioning devices and seats were not used during intervention in order to allow C to use active and self-generated movements. C's extremities and body were not passively moved, objects were not placed in her hands, and her weight was not shifted for her. According to C's mother, the START-Play intervention was very different from her early intervention therapy. C's mother reported she had been trained to provide daily passive range of motion, work on rolling supine to prone moving C's arm or leg across her torso, and to practice prone every day.

Analysis:

Graphic representation of the data and visual inspection were first used to determine if the baseline phase was stable. [23,24] The 2 Standard Deviation (SD) Band Method was used to determine if there was a change in the outcome measures during the intervention phase compared to the baseline. The mean of the baseline phase +/− 2 SD was calculated. Each data point during the intervention and follow-up phases that was more than 2 SD above the mean was considered to represented a significant increase from baseline.

In addition, a second analysis was completed using the Percent Non-Overlapping Data (PND) to compare the baseline and intervention phases. The highest value for each measure during the baseline phase was identified.[24] The percent of data points in the intervention and follow up phases that are higher than the highest baseline measure were calculated and reported as the percent of non-overlapping data (PND). A PND less than 50 was considered to represent no observed effect, 50 to 70 a questionable effect, and more than 70 an intervention effect. [24]

Results

C completed all planned assessment and intervention sessions without adverse events. Using the 2 SD above the baseline method, C's cognitive skills on the Bayley-III improved by the end of the follow up (Table 1, Figure 1). Her gross motor skills on the Bayley-III improved on 100% of the outcome assessments including immediately post-intervention and retention in follow up. Using the PND method, C's cognitive and motor scores on the Bayley-III increased on 67% of the outcome assessments. These results support the conclusion that the START-Play intervention likely contributed to the improvement in this child's cognitive and motor outcomes as measured on the Bayley-III.

On the secondary outcomes, C showed an increased rate of EPSI behaviors, increased GMFM total and sitting scores, and increased frequency of contact with toys during reaching at 100% of outcome assessments using both analysis methods (Table 1, Figure 2). These findings support the conclusion that C made significant motor and problem-solving gains during the intervention and retained or increased these gains following the START-Play intervention.

C's mother increasingly provided cognitive opportunities during and following intervention (Table 1, Figure 2). Prior to the intervention, C's mother used toys to motivate C to try motor skills or to entertain her during passive movements. After the intervention she incorporated the toys into problem solving activities to motivate C to move while exploring the function of the toy. These findings suggest C's mother learned and incorporated the key components of START-Play intervention into free play with C.

Discussion

The results of this study demonstrate that C increased her motor and cognitive skills from the baseline period to follow-up. While it is likely that C would have continued to have a gradual increase in her scores without the intervention, the rate of improvement appears to have increased during and following the intervention. For example, during the 10 months between the Bayley-III baseline assessments, she gained 4 raw points in the cognitive domain (or about 1 new item every 2.5 months. In contrast, during the 3 months of intervention she gained 3 raw points, or 1 item every month. From the end of intervention until 9 months later, C gained 2 raw points, or 1 item every 4.5 months. Both the Bayley-III and the EPSI showed an increase in skill during the intervention with the rate of learning slowed when intervention ended. The lack of consistent gains on the EPSI after the intervention may represent a regression toward the mean, reduced opportunities to practice the problem solving behaviors, or that she was completing fewer but more complex problem solving behaviors that are not reflected in the EPSI scoring.

The motor gains in this study were consistent between all 3 measures: the Bayley-III, GMFM, and reaching. All showed large gains during the intervention that were retained in the long term but did not increase as quickly following intervention. In addition, C's mother changed her interactions increasing the cognitive opportunities provided during the free play assessment. Interestingly, even with the mother's change in behavior to provide increased cognitive opportunities, the increased rate of gains in motor and cognition did not continue during the 9 month follow-up period. We suspect that without the regular intervention visits the mother may not have been aware of how to advance the activities and provide the just-right challenge as C learned.

Taken together, there is a probable increase in the rate of gaining cognitive and motor skills during the intervention period when compared to the baseline period, and the retention of the newly learned skills supports the potential for the START-Play intervention to help advance cognitive and motor skills in infants with severe motor impairment prior to the onset of prop sitting. This unique physical therapy intervention establishes cognitive play as the primary focus of the intervention. While some would consider this outside the realm of physical

therapy, we suggest that cognitive and motor systems should not be treated separately as they are highly intertwined and develop concurrently. In addition, this study provides initial evidence that even with a focus on cognitive function during intervention, motor skills improved considerably. Thus physical therapists should considered that incorporating cognitive tasks with motor tasks may increase the efficacy of intervention by "opening the child's eyes" to new and exciting ways to play, advancing the quality of interaction with people, and exploring the impact of their action/movement on the world.

The findings from this single subject study are consistent with the findings from several other studies that use a similar theoretical model to provide intervention for children with less severe motor impairments.[4,18] Perception and action are required for both motor and cognitive development. Thus, interventions which blend these approaches to target the skills just beyond the child's current level of function are consistent with today's widely accepted theoretical models of development. [13]

This paper supports the need for additional evaluation of the efficacy of START-Play and similar interventions in infants with severe motor impairment. The use of a single subject, lack of a control group, and limited number of data points on the primary outcome during each phase limit our ability to fully quantify the efficacy of the intervention or relate the finding to other children. In addition, the inability to truely withdraw the intervention, since the mother's skills were changed, prohibited the use of a more rigorous ABA design. In addition, while the majority of the outcome measures had stable baseline assessments, there was some variability in the baseline measures that may have resulted in fewer visits being described as different than baseline thus under-reporting the impact of the intervention on the outcomes.

The results of this study must be viewed within the context of this single subject and the purpose of this study. This is the beginning of a course of research needed to evaluate if the START-Play intervention is appropriate for children with more severe motor impairments than it was initially designed for. The purpose of this study was not to make a statement on efficacy, rather to identify the need for research to evaluate efficacy.

Conclusions:

The results of this study suggest that the START-Play intervention has promise for improving motor and cognitive outcomes in children with severe motor delays initiated prior to or in the absence of ability to prop sit for 3 seconds. The targeted intervention to advance sitting, reaching, and problem solving appears to be associated with an increased rate of development of gross motor skills. Improved gross motor skills in combination with increased cognitive opportunities may have contributed to C's cognitive gains. Therapists treating this population should not hesitate to add a focus on cognitive skill into the motor intervention they provide. Working with parents to change the opportunities they provide to support development of the child's self-generated actions is likely to lead to gain in multiple domains. Additional research on the efficacy of the START-Play intervention in children with severe motor impairments before the onset of prop sitting is warranted.

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DOCUMENTATION

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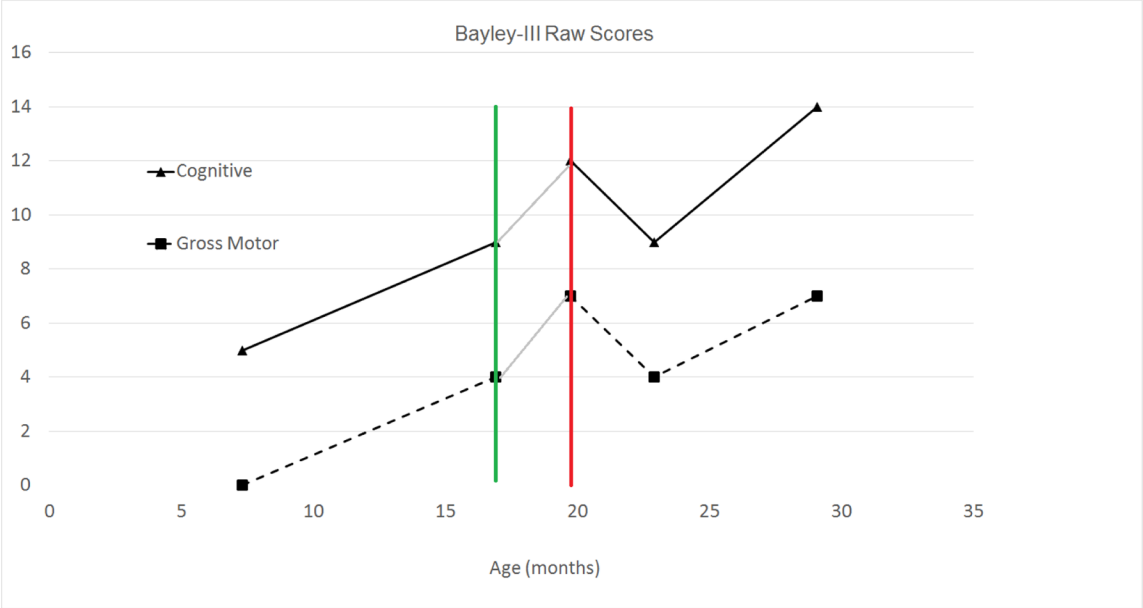

Figure 1:

Raw Scores on the Bayley Scales of Infant and Toddler Development Motor and Cognitive Scales. The green line represented the start on intervention. The red line represented the end of intervention. The light gray line indicates the change from the beginning to end of intervention.

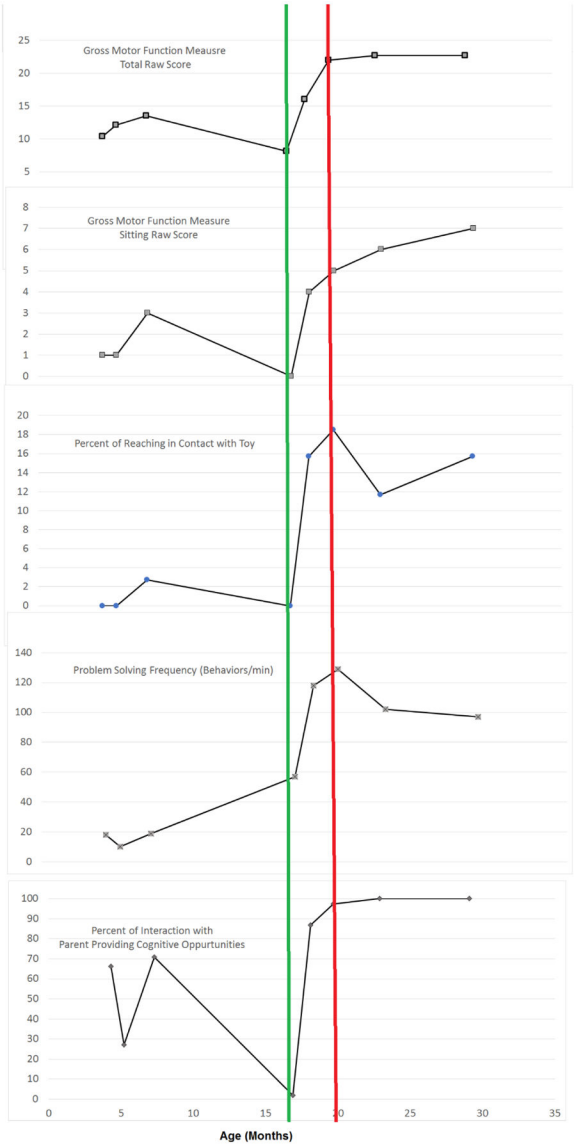

Figure 2:

All secondary outcome measures at each data point available. The vertical green line represents the start and the vertical red line represents the end of intervention.

Table 1:

This table presents the raw data for C. The shaded columns represent the intervention phase.

	Baseline 1	**Baseline 2**	**Baseline 3**	**Baseline 4- start intervention**	**Intervention 1 – mid intervention**	**Intervention 2 – end intervention**	**Follow -up 1**	**Follow – up 2**	**% of the visits meeting 2 Standard Deviation Criteria**	**% of the visits meeting Percent Non-Overlapping Criteria**
Age (months)	4.3	5.2	7.3	16.9	18.1	19.73	22.7	29.1		
Bayley Cognitive Raw Score	NA	NA	5	9	NA	12[+]	9	14[*+]	33	67 questionable effect
Bayley Gross Motor Raw Score	NA	NA	0	4	NA	7[*+]	4[*]	7[*+]	100	67 questionable effect
GMFM 66 - IS	10.4	12.1	13.5	8.1	16.0[*+]	22.0[*+]	22.7[*+]	22.7[*+]	100	100 intervention was effective
GMFM SS	1	1	3	0	4[*+]	5[*+]	6[*+]	7[*+]	100	100 intervention was effective
Reaching frequency of all contact	0	0	3	0	16[*+]	19[*+]	12[*+]	16[*+]	100	100 intervention was effective
Total Rate of EPSI behavior	3.0	1.7	3.2	9.5	19.7[*+]	21.5[*+]	16.8[*+]	16.2[*+]	100	100 intervention was effective
% Cognitive opportunities	66.3	27.2	70.8	2.0	86.9[+]	97.4[+]	100[+]	100[+]	NA	100 intervention was effective

[*] The values in the intervention phase and follow up which were more than 2 SD above the baseline mean are indicated with an.

[+] The values in the intervention phase and follow up that were over the highest baseline value are indicated with a.

The percent of the intervention and follow up visits that meet the 2 SD and PNO are listed for each outcome. If the percent could not be mathematically determined it is marked as not applicable (NA).