Exploring Effects of the HEP® (Homeostasis-Enrichment-Plasticity) Approach as a Comprehensive Therapy Intervention for an Infant with Cerebral Palsy: A Case Report

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Case Report

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Posted Date: April 11th, 2022

DOI: https://doi.org/10.21203/rs.3.rs-1524867/v1

Exploring Effects of the HEP® (Homeostasis-Enrichment-Plasticity) Approach as a Comprehensive Therapy Intervention for an Infant with Cerebral Palsy: A Case Report

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Abstract

Background: Cerebral palsy (CP) is a common non-progressive neurodevelopmental disorder which causes developmental disabilities in children. Varied interventions for CP exist to address medical and physical needs but with limited effectiveness evidence. Environmental Enrichment (EE) is an animal model intervention for many neurodevelopmental disorders, including CP, with considerable positive effects. This case report defines the "HEP®" (Homeostasis-Enrichment-Plasticity) approach, which is based upon principles of EE and ecological theories of development, and describes its' use to promote the developmental and functional skills of an infant with CP.

Methods: Parent interviews and assessment data were completed before and after intervention. Data was gathered by developmental history, systematic observation of behaviors in the clinical setting and at home, Beck Anxiety Inventory (BAI), Infant-Toddler Symptom Checklist, the Sensory Profile Infant/Toddler, Peabody Developmental Motor Scales-2, Gross Motor Function Measurement-88 (GMFM-88), the Gross Motor Function Classification System (GMFCS) and Pediatric Evaluation of Disability Inventory (PEDI). The HEP® approach intervention was implemented one time per week for 12 months.

Results: Following the HEP® approach intervention, self regulation and sensory processing scores improve to typical performance ranges. GMFM-88 total score improved from 45/264 to 123/264. The Peabody found all Gross Motor (54 to 110), Fine Motor (65 to 117) and Total Motor Quotient (119 to 227) scores increased significantly after intervention**.** Post-intervention observations showed significant gross motor progress with movement from

GMFCS Level IV to Level I. Performance on the Functional Skills Scales and Caregiver Assistance Scales of PEDI also demonstrated significant gains.

Conclusions: A definition and detailed description of the HEP® approach intervention is presented here for the first time. The case report demonstrated preliminary evidence for the effectiveness of the HEP® approach on self-regulation, sensory processing, motor development, functional skills, and caregiver assistance with an infant with CP. Additional studies are needed to validate the findings.

Keywords: Enriched Environment, Cerebral Palsy, Physical Therapy, Early Intervention

Introduction

Cerebral palsy (CP) is a non-progressive neurodevelopmental disorder that results from lesions occurring in the developing infant brain. Although CP is traditionally described as a disorder of movement and posture development that causes activity limitations, more recent definitions allow clinicians to appreciate that CP may also affect sensation, perception, cognition, communication, and behavior (Wimalasundera and Stevenson, 2016; Richards and Malouin, 2013; Rosenbaum et al., 2006). The prevalence of CP worldwide is 2.11 per 1000 live births and has remained constant in recent years (Oskoui et al., 2013). Up to the last decade, interventions for CP have predominantly focused on medical and physical needs, often with limited evidence to support their efficacy (Wimalasundera and Stevenson, 2016). However, evidence-based interventions for CP that provide clinicians and families newer, safer, and more effective possibilities have continued to expand in recent years (Novak et al., 2020) A review by Novak et al. (2020) found that considerable clinical trial data supported the efficacy of training-based interventions (i.e. action observation training, bimanual training, constraint-induced movement therapy, goal-directed training) for motor impairments and difficulties with tasks involving motor performance. Recently, there has been increased interest in environmental enrichment-based interventions and their potential therapeutic benefits in many neurodevelopmental disorders, including CP (Ball et al., 2019).

Moreover, Novak et al. (2020) pointed to evidence supporting environmental enrichment as an effective intervention for promoting task performance in children with CP.

Environmental enrichment (EE) is a paradigm that emerged from experimental studies on animals (predominently mice) and has been described as manipulation of standard laboratory conditions that modify the quality and intensity of environmental stimulation. EE interventions provide increased levels of multisensory stimulation, physical activity and social interactions through eliciting spontaneous explorative behaviours (Baroncelli et al., 2010). While there are varying EE protocols, the key features of EE are: 1) large spaces that induces active exploration; 2) variety of objects (i.e. objects with different size, shape, weight, and texture, climbing settings, tubes or tunnels, balance platforms, mazes, running wheels and balls that facilitates cognitive, sensory and motor experiences) that are changed or differently oriented for novelty, challenge, adaptability and complexity, and 3) multiple subjects for increased socialization opportunities. Other basic features, which are not usually stated but are common in EE studies, are the provision of homeostatic needs such as food, water, heat, sleep cycles and health conditions, and the continuation of EE conditions for a extended period (Nithianantharajah and Hannan, 2006; Reynolds et al., 2010; Slater and Cao 2015; McCreary and Metz, 2016). Despite inconsistency in the specific composition of EE (i.e. the cage size, group size, toys, tasks, materilas, duration, etc.) in varied experiments, EE research has consistently demonstated positive outcomes in motor performance, socialization and learning(Kempermann, 2019).

Improvements following environmental enrichment are attributed to active interaction between the individual and the affordances available in the environment. Over half a century of research has showed that EE positively facilitates neuronal activity, gene expression, epigentic modification, signaling factor, neurotransmitter level, neurotransmitter receptor expression, cortical thickness, brain weight, disease phenotype, dendritic morphology, spine formation, synaptic plasticity, adult neurogenesis, learning and memory, affective behavior and resistance to stress (Kempermann, 2019; Diamond et al., 1966; Rosenzweig and Bennett, 1972; Van Praag et al., 2000; Sale et al., 2012; Alwis and Rajan, 2014; Sampedro-Piquero and Begega, 2017). In particular, a considerable number of studies have shown that EE leads to improvement in neurodevelopmental impairments and behavioral deficits that occur as a result of brain damage early in life (Rojas et al., 2013; Marques et al., 2014; Schuch et al., 2016; Griva et al., 2017; Durán-Carabali et al., 2018; 2019a; 2019b). Morever, it is suggested that EE may be used as a non-invasive and non-pharmacological intervention against various neurological condition such as Parkinson's disease, amyotrophic lateral sclerosis, fragile X syndrome, Down syndrome and various other forms of brain injury (Nithianantharajah and Hannan, 2006; Jain et al., 2013; Forbes et al., 2020).

There are emerging interventions inspired by EE for adults with neurological conditions in recent years (Janssen et al., 2014; Rosbergen et al., 2017) and children (Morgan et al., 2016; Dusing et al., 2018). These are more appropriately defined as “enriched therapy” (ET) as they provide limited enriched experiences and do not include all core aspects of an EE paradigm. ET involves therapist-provided stimuli for a specified period of time (usually daily) under certain conditions, whereas, EE involves sensory/environmental adaptations that offer continuous opportunities in social, sensory, motor and cognitive areas through spontaneous exploration (Natali et al., 2020).

In light of these considerations, the “HEP®” (Homeostasis-Enrichment-Plasticity) intervention approach was developed based on principles of EE and ecological theories of development. The HEP® approach applies the core principles of enriched environment paradigms and neural plasticity used in experimental animal studies, in the context of ecological theories of human development, and emphasizes the fundamental importance of homeostasis in the client. Although the approach has a strong theoretical basis, investigation of its clinical results is needed to encourage further development of the model. For this purpose, a case report is presented as a first step in examination of the clinical application of this theoretical model.

The aim of this case report was to define the HEP® approach and explore preliminary effectiveness of the HEP® approach intervention on the regulatory capacity, sensory processing, motor development and functional abilities of an infant with cerebral palsy.

Method

Study Design

A descriptive case report design examined effects of the HEP® approach with an infant diagnosed with CP. While case reports have limitations, they represent an important study design for introducing new, innovative, clinical approaches and to advance scientific knowledge (Carey, 2010). This approach provides the researcher an opportunity to collect data from various sources; to analyze data to illuminate the case (Baxter and Jack, 2008; Heale and Twycross, 2018); and to examine preliminary intervention outcomes (DePoy and Gtlin, 2020). Although the case report approach does not allow generalization of findings, it informs clinical practice by explicating clinical problems and useful solutions.

A systematic method was utilized to gather information and organize data. Written consent was obtained from the family of the individual participating in the case study before initiation of the study. The child's initials were changed to maintain confidentiality.

Participant

YZ was a 12 month (9 months adjusted age) old female with Cerebral Palsy born prematurely at 27 weeks of pregnancy at 970 grams of weight in Istanbul, Turkey. YZ's family requested a second opinion evaluation from the author due to slower than expected motor gains from the child's hospital-based physical therapy. A detailed developmental history gathered from the family revealed that YZ had a grade 4 brain bleed immediately after birth, remained in the intensive care unit for 3 months and received respiratory support for a considerable period of time. Symptoms of hydrocephalus appeared soon after discharge, and the infant subsequently underwent endoscopic brain surgery. The infant received treatment for retinopathy of prematurity as well. When medical conditions were stabilized a developmental assessment was completed at the hospital. As a result of the evaluation, delayed motor development and increased muscle tone in the left upper extremity was identified. Subsequently, the infant received physiotherapy in the hospital until the age of 12 months.

Primary parental concerns were delayed motor development and limited use of the left upper extremity. Parents stated that YZ's age-appropriate gross motor skills such as rolling, creeping on belly or four-point crawling, sitting, standing, and positional transitions (from lying to sitting or from sitting to standing) were not yet developed. Further, they claimed that the infant's left hand was often closed, bent at her side, and that she did not use this hand actively in any way. In addition, the family reported that the infant was fussy and cried often during the physiotherapy sessions, so they had difficulty doing exercises both in the clinical environment and at home.

Assessments

In addition to the detailed parent interview and informal observations of the child, an independent evaluator completed a battery of assessments before and after intervention. Before intervention, a developmental history, interview with the parents, systematic observation of behaviors in the clinical setting and at home, Beck Anxiety Inventory (BAI), Infant-Toddler Symptom Checklist (ITSC), the Sensory Profile Infant/Toddler (SPIT), Peabody Developmental Motor Scales-2 (PDMS-2), Gross Motor Function Measurement-88 (GMFM-88), the Gross Motor Function Classification System (GMFCS) and Pediatric Evaluation of Disability Inventory (PEDI) were completed. Post intervention documentation was collected at the end of 12 months of intervention. A parent interview was also conducted post intervention to obtain input about the child's past and present concerns and to investigate the parent's perception of the HEP® approach intervention program's success in meeting their child's needs.

The Beck Anxiety Inventory (BAI) examined parental anxiety. BAI is a 21- item self-report questionnaire that measures the frequency of ones experience of anxiety symptoms. Each item provides Likert type (0=none, 3=intensive) measurement over four points (Beck et al., 1988). The total score ranges from 0 to 63 and has been found to be valid and reliable in a Turkish population (Ulusoy e al., 1998).

The Infant-Toddler Symptom Checklist (ITSC) examined self-regulation. ITSC is used from 7 to 30 months of age and focuses on the infant's responses in the domains of self-regulation, attention, sleep, feeding, dressing, bathing, and touch, movement, listening, language, and sound, looking and sight, and attachment/emotional functioning. A criterion-group validation model was used and optimal cutoff scores were located. Infants scoring at or above a cutoff score in any category were considered "at risk" (DeGangi et al., 1995; DeGangi, 2000; DeGangi et al., 2000).

The Infant/Toddler Sensory Profile (ITSP) examined sensory processing. ITSP is a 48-item caregiver questionnaire that measures sensory processing abilities in children ages 7 months to 36 months. Frequency of child behaviors are rated by parents on a 5-point likert scale (1=almost always, 5=almost never). The total frequency of behaviors for Auditory, Visual, Vestibular, Tactile, and Oral Sensory modulation is calculated individually. Scores are then grouped into four quadrant scores: Low Registration, Sensation Seeking, Sensory Sensitivity, and Sensation Avoiding. A low threshold score is calculated by summing Sensitivity and Avoiding quadrant scores. Lower scores indicate a higher frequency of response. Reliabilities for the various composite scores ranged from .69 to .85. Test validity was established in several studies (Dunn, 2002).

PDMS-2 and GMFM-88 measured motor abilities, and the GMFCS classified level of gross motor function. The PDMS-2 is comprised of three gross motor and two fine motor subtest. Scoring of the PDMS-2 generates a raw score for each subtest, individual subtest scale scores, and an overall gross motor and fine motor quotient. The PDMS-2 has good discriminative reliability and validity and test-retest reliability is also high (Folio and Fewell, 2010).

The GMFM-88 monitored progress in gross motor development. GMFM-88 is a standardized criterion referenced measure designed to be used for both clinical and research purposes to measure change over time and effectiveness of interventions for children with disabilities, ages 5 months to 16 years of age (Russell et al., 2002; Alotaibi et al., 2014; Salavati et al., 2015).

GMFCS examined self-initiated functional movements, with emphasis on head and trunk control, sitting, transfers, and mobility for CP. The GMFCS is a five-level pattern-recognition system (Level I represents the best gross motor abilities and Level V the least function). The classification of level-specific motor abilities is age dependent, and there are specific motor abilities definitions for various levels in each age band. There are five described age bands: before the second birthday, from age 2 to 4, from 4 to 6, from age 6 to12 years, and from age 12 to 18 (Palisano, 1997).

The PEDI evaluated the functional skill performance and necessary assistance and modifications in children aged 6 months to 7.5 years (Haley et al., 1992). It includes three sets of scales: Functional Skills, Caregiver Assistance, and Modifications (197 functional skill items, and 20 items for caregiver assistance and modifications). The Functional Skills Scale (FSS) provides sufficient detail to identify clinical patterns of deficiences in functional skill attainment, while Caregiver Assistance Scales (CAS) measures actual performance by the extent of help a parent gives in daily functioning. The CAS provides additional information to the results of the FSS, in fact the CAS is an indirect measure of capability, whereas the FSS is a direct measure. The Modifications Scale (MS) is the assessment of environmental modifications that support functional performance (Wassenberg-Severijnen e al., 2006). The PEDI is valid and reliable tool for the Turkish population (Erkin et al., 2007).

Intervention

Theoretical Framework

The theoretical model of the HEP® approach articulates factors affecting behavior and development in a basic framework (Fig.1). It also guides assessment and hypothesis generation for intervention. This framework was developed based on ecological theories of development including Ecological Theory, Dynamical Systems Theory, Perception-Action Theory, Theory of Neuronal Group Selection, and Person-Environment-Occupation Model (Bronfenbrenner, 1992; Thelenand Smith, 1994; Newell, 1986; Gibson, 1979; Edelman, 1987; Law et al., 1996).

The model consists of four main sections: 1) *Environment:* This part of the model highlights effects of the social and physical environment on development. It derives from theories that discuss effects of the environment on behavior and development (Bronfenbrenner, 1992; Newell, 1986; Law et al., 1996; Shumway-Cook and Woollacott, 2017); 2) *Time:* Effects of temporal factors on development are articulated in this section (Bronfenbrenner, 1992). Here the importance of past and present experiences and expectations for the future are emphasized; 3) *Task:* Although researchers define the task section as necessary body functions (Shumway-Cook and Woollacott, 2017) or goals, rules and tools of the task (Newell, 1986) in this model we determine if the task is in the child's "zone of proximal development" (Vygotsky, 2012) and if it is meaningful for the individual (Edelman, 1987); *4) Individual:* Five key individual variables affecting an individuals unique pattern of behaviors or development have been identified (adapted from Connolly & Montgomery, 2001): Homeostasis, sensation/sensory processing, emotion, motor and cognition are considered intrinsic control parameters that influence the child's behavior and development in a non-liner fashion (Thelen and Smith, 1994). *Homeostasis* refers to an individual's general health status (e.g. constipation, allergies, reflux, weight gain, growth, medication, seizures, etc.), sleep hygiene, arousal and stress level (DeGangi, 2000; 2017). *Sensation/Sensory processing* is the ability of an individual's brain and nervous system to register, modulate, discriminate, perceive, and interpret sensory information to generate an adaptive response

(Ayres, 2005; Bundy and Lane, 2020). *Emotion* includes the individual's functional emotional developmental level (Greenspan et al., 2001), attachment style (Ainsworth et al., 2015), trauma history and general mood. Musculoskeletal constraints (e.g. flexibility, range of motion, muscle tone, strength), postural control (e.g. balance, stability) and motor skills (e.g. roll, crawl, walk, etc.) are the *motor* variables that contribute to development (Connolly and Montgomery, 2001). Lastly, *cognition* consists of attention, language, problem solving, planning, social thinking and so on.

Figure 1. Theoretical framework of the HEP® approach.

Intervention Principles

The intervention principles of the HEP® approach were developed from essential features of EE: physiological homeostasis, safety, sensory experiences, spatial features of the environment, environmental and object novelty, challenge, enjoyment, continuity, social opportunities, active engagement in and exploration of the environment (Nithianantharajah and Hannan, 2006; Reynolds et al., 2010; Slater and Cao, 2015; McCreary and Metz, 2016). In the HEP® approach, these key features of EE are implemented with the guidance of core principles of Dynamical Systems Theory, Gibson's Ecological Theory of Perception, Theory of Neuronal Group Selection, Polyvagal Theory, and Synactive Theory. Table 1 presents definitions of the various specific aspects of the HEP® approach and provides examples of how these principles were implemented with the case of YZ.

Table 1.

HEP® approach intervention principles.

Key Features of HEP® intervention	Description for Implementation of Intervention Principles	YZ Case ExampleActivities
Physiological Homeostasis	Achieving and maintaining homeostasis is the primary goal of a living organism. It is described as regulating internal environmental conditions according to changes in the external environment for health and functionality. It is the dynamic balance of sympathetic and parasympathetic systems and is a prerequisite for active exploration, learning and development. In this model, the therapist prioritizes homeostasis while considering how all developmental areas will be affected by changes in homeostatic states (Als, 1982; 1986; Mouradian and Als, 1994; Schulkin, 2004; Brezelton and Nugent, 2011; Porges, 2011; Guyton and Hall, 2011; Maltese et al., 2017)	YZ's family was supported to implement home routines that affected YZ's homeostasis in areas such as sleep and nutrition. Parents were counseled in the areas of the child's self-regulation and importance and methods of co-regulation. The importance of active movement of the baby for self-regulation was emphasized and suggestions such as use of a baby jumper were provided.
Safety	To interact actively and effectively with the environment an individual's nervous system must perceive itself as in a place and state of physical and emotional safety. Perception of security is the foundation for active exploration and participation necessary for learning and development. The therapist supports the perception of security by collaborating with the individual and adapting the environment (Porges, 2011; Perry and Szalavitz, 2017; Schore, 2015).	Strategies were suggested to the family for supporting the child's perception of physical and emotional safety. For physical safety adequate space and materials such as a basket sitting or a hollow cylinder for standing were provided. With these adaptations the child was able to actively move and explore without fear of falling. For emotional safety, the parents informed the child about changes in the environment in advance or made transitions slower thus facilitating the baby's safe adaptation to the environment.
Sensory Experiences	Perception of sensory information and subsequent production of adaptive responses during sensory experiences are fundamental sources of learning and development. Robust sensory systems facilitate perception and act as control parameters (factors) for new actions. Therapists support perception through use of activities that emphasis the child's strong sensory systems to achieve actions that are meaningful to the individual (Thelen and Smith, 1994; Edelman, 1987; Ayres, 2005; Bundy and Lane, 2020; Adolph et al., 1993; Adolph, 2008; Adolph et al., 2019; Gibson, 2015).	The importance of vision and hearing(which were the child's strong sensory systems) for moving, exploring and interacting was explained to the parents. To support the use of vision and hearing in active exploration and movement, vertical positions with the baby's body parts within the visual field were suggested.
Spatial	Physical characteristics of the child's space (such as the size of the space, support surfaces in the vertical and transverse plane, objects and equipments that facilitate active movement or exploration) are parameters that have the capacity to significantly affect behavior. When the space (support surfaces provided via equipments) provides support in the zone of proximal development, it will facilitate active exploration in a a wider space (the explored space will increase). The therapist provides spatial conditions (such as box for sitting or standing) that are	Spatial conditions suitable for the infant's capacity were provided to support the infant's active movement and exploration. For example, a cardboard box was provided that closely supported the torso so the baby could actively explore mobility skills required for sitting; and free hands to engage in interaction potentials in the environment. Later, as the infant's capacity developed, physical supports were provided by a large inner tube , which gave the infant more space for active exploration and

	tailored to the child's individual profile to support spontaneous self-organization, stimulate perception, encourage action and promote active exploration. As the environment that is actively explored expands development is facilitated (Bronfenbrenner, 1992; Thelen and Smith, 1994; Newell, 1986; Adolph, 2008; Adolph et al., 2019; Gibson, 2015).	movement. In a standing position the infant was initially supported with a baby jumper which provided total support and encouraged exploration of active movement. Later as the baby developed her capaticity for active movement and exploration the support was decreased and the space enhanced; e.g. standing and moving inside small tires, followed by moving inside bigger tires, and then moving inside a cardboard made channel, and continued with hand supported side walking beside the furniture in the living room and with cardboard boxes lined up along the walls in the whole house.
Novelty	Novelty is an important feature for developmental change and facilitation of adaptability. The therapist changes or adjusts the environment to stimulate new action affordances. The novelty presented should be noticed by the individual, stimulate perception, and result in action. Every novelty should be built as a new variation on existing neuronal models/schemas (Thelen and Smith, 1994; Newell, 1986; Edelman, 1987; Adolph et al., 1993; Adolph, 2008; Adolph et al., 2019).	Changes were routinely made to the above activities to add novelty to the active experiences with which the baby was familiar. For example, to change the direction of the baby's movement, the location of toys and the direction of the equipment relative to the baby's position were changed, different equipment (such as a basket instead of a box) that offered similar experiences or supports were used, or familiar games were played with different objects. These changes were presented after the infant could easily accomplish and adapt to an earlier experience.
Challenge	Challenge is a natural stimulant of development and encourages complexity. Learning emerges when the challenge is adjusted to the child's zone of proximal development. Once the individual has developed a skill or behavior, variations of that skill are the new challenges the individual needs to adapt to. Variations lay the groundwork for more complex skills. All systems spontaneously self-organize to meet the new challenge (Thelen and Smith, 1994; Edelman, 1987; Vygotsky, 2012; Ayres, 2005; Adolph et al., 1993; Adolph, 2008; Adolph et al., 2019; Gibson, 2015).	As the baby's capacity developed, challenges were presented that were in her developmental ability. The size of the space for movement while sitting and standing or the distance the baby could travel was increased as the capacity of active exploration and movement developed.
Enjoyment	Learning emerges in activities that are meaningful, purposeful and motivating for the individual. Enjoyable active experiences (sensory, motor, social, cognitive) in the zone of proximal development have an adaptive value and stimulate new behaviors or skills (Thelen and Smith, 1994; Edelman, 1987; Vygotsky, 2012; Ayres, 2005).	The importance of the child's motivation and fun in learning and development was explained to the family. Activities that motivated the baby and were meaningful for her were chosen. The baby was highly motivated by books and symbolic play, and therefore her movement was encouraged to engage in activities that were meaningful to her (e.g. symbolic play and book that were meaningful for the baby were used as part of the activity).
Continuity	To acquire the benefits of an enrichment paradigm, exposure to EE should be ongoing. Family and other caregivers must be at the center of the intervention so that enrichment strategies can be applied in every moment of life. The fact that these strategies are acceptable, accessible and sustainable support continuity of the intervention (Kempermann, 2019; Harland and Dalrymple-Alford,	The importance of continuity for the enriching effects of environmental adaptation was explained to the family. For this education, live and online parental consultation was provided on how enrichment practices should be done. Activities were organized in a way that would be everywhere in daily life, in harmony with the household routines and rituals and

	2020).	the family's own dynamics. For this, materials that can be found everywhere, such as cardboard boxes, baskets, car tires, and activities that can be easily provided by any caregiver were suggested.
Social	Social experiences available to the individual within their zone of proximal development provide enriching effects. The social environment consists of every adult and child around the individual and provides important stimulus for development. Scaffolding of the individual by more knowledgeable others in the zone of proximal development facilitates learning (Bronfenbrenner, 1992; Vygotsky, 2012; Greenspan et al., 2001).	Interaction strategies appropriate to the baby's capacity were taught to all family members and continuous counseling was provided in this regard. For example, it was explained that while communicating with the baby, less words, and sentences, while more gestures and mimics should be used. In addition, it was recommended to utilize environments that offered novel social interaction opportunities for the baby. For example, more frequent visits to neighbors and relatives and promoting interaction with new people were recommended. Effective scaffolding strategies were demonstrated to the family so that the baby could better interact with new people in new environments, e.g., mother explained behaviors of others to the baby, and then baby's behaviors to others.
Active Engagement and Exploration	To achieve the effects of EE, the individual must actively explore the possibilities of acting on the environment. The physical and social environment should encourage and support the child's spontaneous active exploration and engagement. Physiological systems have the capacity to spontaneously self-organize for active exploration. Developmental changes of behaviors emerge with spontaneous self-organization of systems, and this requires different time for different individuals (Bronfenbrenner, 1992; Thelen and Smith, 1994).	Parents were taught that the people around the baby and the space or the equipment provided should encourage the baby's active exploration and that she should be given enough time for this exploration. For example, if the baby could not stand with support from furniture while she could stand inside the tires, the tires were accepted as the space and equipment that supported active exploration and participation. Also, baby jumper, baby walker and infant ride on car were identified as equipment that facilitated active exploration. On the other hand, the adults around the baby were instructed to facilitate active exploration by providing effective reinforcers such as gestures, toys or appropriate play opportunities.

Based on YZ's evaluation data, a hypothesis was generated regarding possible factors underlying her functional challenges. Intervention goals were established with the parents to reflect their areas of concern. Intervention was then provided for 75-minute sessions (45 minutes child-focused session and 30 minutes parent interview), one session per week, over a period of 12 months. With the guidance of the HEP® intervention principles, the therapist first addressed homeostatic problems and collaborated with the family for home carry over activities. For example, the therapist suggested massage and co-regulation strategies to the parents to support the child's self-regulation. Second, the importance of physical and emotional safety for the child and the core principles for supporting the child's perception of safety were explained to the caregivers. Next, the therapist provided the parents with home activity suggestions in accordance with the other intervention principles such as creating spatial supports, novelty, challenge in the zone proximal development and facilitating enjoyable active exploration and sensory experiences in a continuous way (see Fig. 2 and Fig. 3 for examples of home activity progression over time). Implementation of these suggestions was checked in terms of parent implementation of challenge, novelty and continuity principles of the intervention regularly between sessions via parent-provided video.

Figure 2. HEP® approach home application examples. Photographs represent the temporal progression of the intervention. Spatial supports that facilitated active exploration and sensory experiences through meaningful activities were changed over time (from A, age 9 months, to G, age 21 months adjusted) according to novelty and challenge principles of the HEP® approach. As shown in picture A, the baby's active exploration, sensory experience and social participation through meaningful and fun activities are provided within a space designed to closely support the body. Picture D shows that the infant's spatial support arrangement has changed and the space for active exploration expanded. Thus, the infant had more sensory experience opportunities through the novelty and challenge provided with the increasing spatial surround. Parents were coached on principles of spatial support and later they created their own spatial support equipments (eg. picture C notes the use of a simple box, paper towels rolls and a dowel to facilitate exploration of active sitting).

Figure 3. HEP® approach home application examples from 9 months to 21 months of age (adjusted). Spatial supports that facilitated active exploration and sensory experiences through meaningful activities, were changed over time (from A to I) according to novelty and challenge principles of the HEP® approach.

Results

Initial observations of YZ were made during free interactions and play with her parents. It was noted that YZ often preferred to sit on her parent's lap, was generally anxious and timid, preferred her right hand for all object manipulation even when asked to use her left hand, and had no interest in new toys. YZ's left arm was often in flexion while with her hand closed and passive.

According to the ITSP parent questionnaire auditory, visual, vestibular, tactile, and oral sensory processing were in the typical range before intervention. Quadrant scores revealed a typical range for low registration (47 points) and sensation seeking (41 points), whereas a probable difference was noted for sensory sensitivity (38 points), sensation avoiding (43 points), and low threshold (81 points). After intervention sensory sensitivity (47 points), sensation avoiding (50 points), and low threshold (97 points) quadrants were in the typical performance range for her adjusted developmental age.

ITSC scores were in the typical range before intervention except for self-regulation and dressing, bathing, and touch subtests. Both, areas of self-regulation and dressing, bathing, and touch areas improved and were in the typical range at post-intervention assessment.

Structured evaluation with the GMFCS revealed that YZ had difficulty with movement transitions (e.g. from supine to prone or sitting position; from sitting to prone or supine position or four point crawling position); pivoting on tummy; crawling on belly and/or four point crawling; independent sitting and bearing weight on the legs while supported in vertical by parents). According to observed performance in gross motor function the infant was initially classified as Level IV on the GMFCS (Table 2). For her age this was equivalent to a developmental level of 4-5 months. After 12 months of intervention YZ was observed to be calm and alert enough to interact with the environment and people. Parents stated that the spontaneous use of the left hand increased in the home environment. Clinical observations showed that her left upper extremity was more active, less flexed and the hand was more open compared to her initial assessment. Mother stated that the infant's interest in play and toys had increased to the extend that YZ now demonstrated symbolic play. Rolling, crawling on belly, transition to sitting (from both supine and prone positions), independent sitting, transition from sitting to standing, crawling on and of 4-5 steps at stairs, side walking with the support of furniture and 5-10 independent steps were observed at the clinical setting. Inspection of videos recorded in the home environment also supported our clinical observations. Post-intervention observations showed the infant had made significant progress with a GMFCS level I (Table 2). This indicated an improvement of 9-10 months of development over 12 months of intervention. This indicated a greater gain than would have been expected by normal development alone, thus supporting the efficacy of the HEP intervention.

Table 2.

Description GMFCS Levels and Infant's motor performance according to GMFCS levels.

GMFCS Levels	Description of Levels	Descriptions Specific to the Case
Level I	Infants move in and out of sitting and floor sit with both hands free to manipulate objects. Infants crawl on hands and knees, pull to stand and take steps holding on to furniture. Infants walk between 18 months and 2 years of age without the need for any assistive mobility device	Post-Intervention: Infant demonstrates all Level I requirements except crawling on hands and knees and walking independently (She take only 5-10 independent steps yet).
Level II	Infants maintain floor sitting but may need to use their hands for support to maintain balance. Infants creep on their stomach or crawl on hands and knees. Infants may pull to stand and take steps holding on to furniture.	

Level III	Infants maintain floor sitting when the low back is supported. Infants roll and creep forward on their stomachs.	
Level IV	Infants have head control but trunk support is required for floor sitting. Infants can roll to supine and may roll to prone.	Pre-Intervention: Infant demonstrated head control while supported by parents for sitting but was not able to roll out of supine or prone before intervention.
Level V	Physical impairments limit voluntary control of movement. Infants are unable to maintain antigravity head and trunk postures in prone and sitting. Infants require adult assistance to roll.	

Pre-intervention GMFM scores were 30/51 for lying & rolling, 15/60 for sitting, 0/42 for crawling and kneeling, 0/39 for standing, 0/72 for walking, running, & jumping and 45/264 for total. By post-intervention, YZ had improved on all GMFM subscores and total GMFM score (Fig. 4). Her scores improved to 48/51 for lying and rolling, 49/60 for sitting, 9/42 for crawling & kneeling, 5/39 for standing, 12/72 for walking, running, and jumping, and 123/264 for total post-intervention. See Figure 4 for pre-post intervention comparisons of GMFM-88 scores.

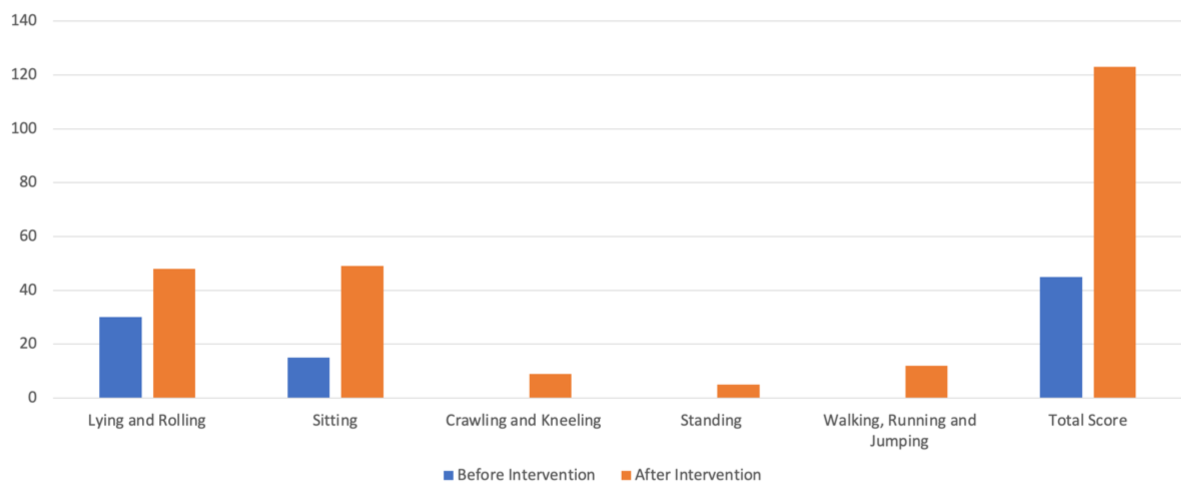

Figure 4. GMFM-88 scores before and after intervention.

The PDMS-2 assessment results revealed that YZ had lower performance in both gross motor and fine motor areas than her age mates before the intervention. Although, YZ did not achieve age-appropriate motor functions after intervention, she demonstrated significant improvements on all subtests of gross motor and fine motor (Fig. 5). Improvements in scores pre- to post-intervention were greater than the confidence interval for each score indicating significant improvement in that area. Improvements noted were reflexes (from 2 to 15), stationary (from 24 to 36), locomotion (from 28 to 57), object manipulation (from 0 to 2), grasping (from 31 to 38) and visual-motor integration (from 34 to 79). All Gross Motor (GMQ), Fine Motor (FMQ) and Total Motor Quotient (TMQ) scores demonstrated significant improvement after intervention (Fig. 6). See (Table 3) for details.

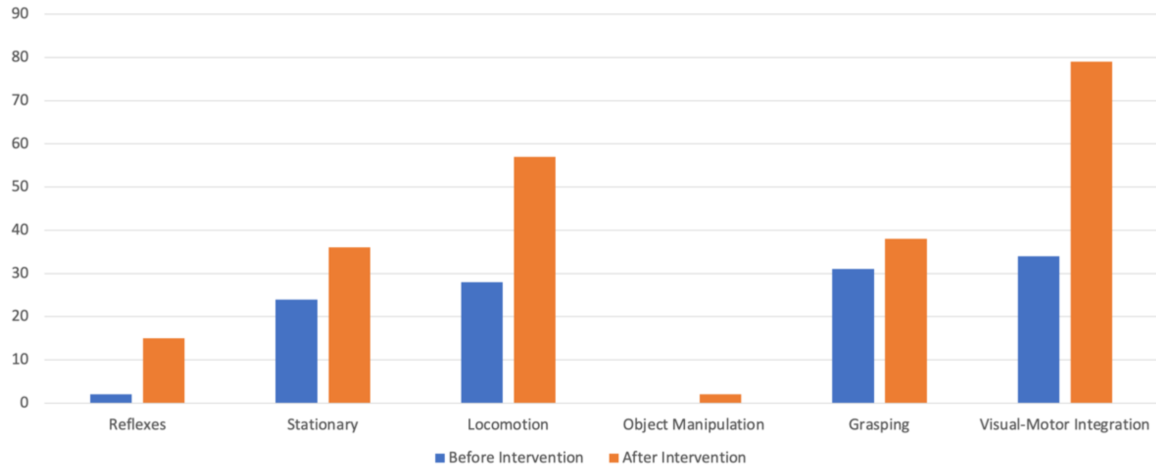

Figure 5. PDMS-2 gross motor and fine motor subtest scores before and after intervention.

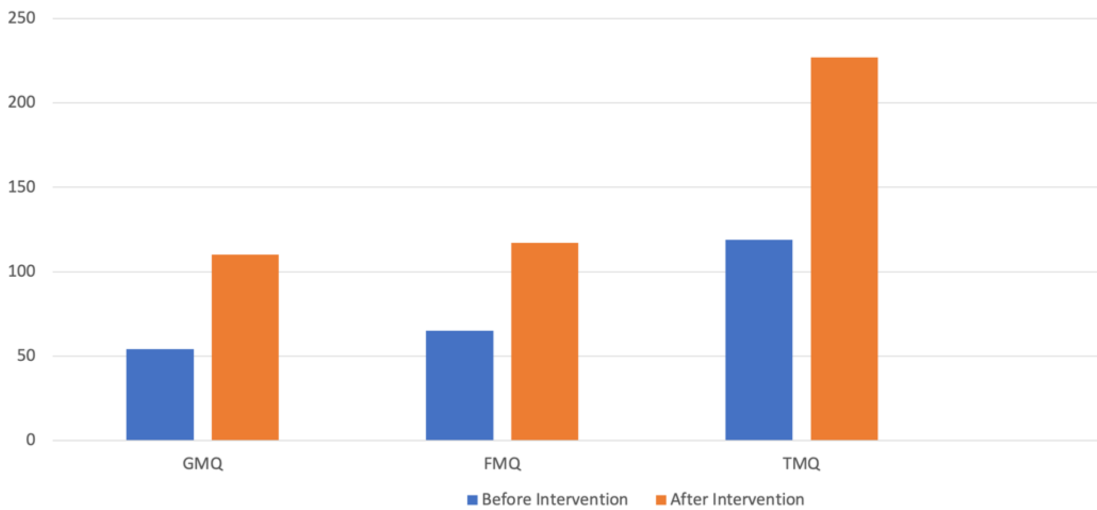

Figure 6. PDMS-2 quotient scores before and after intervention. GMQ=Gross Motor Quotient, FMQ=Fine Motor Quotient, TMQ=Total Motor Quotient.

Table 3.

PDMS-2 Percentile Rank, Quotient Scores, Confidence Intervals and Descriptive Ratings Before and After Intervention.

Quotient	Percentile Rank		Quotient Scores		95% Interval		Descriptive Rating	
	BI	AI	BI	AI	BI	AI	BI	AI
GMQ	＜1	2	61	70	55--67	64--76	Very Poor	Poor
FMQ	5	16	76	85	70--82	79--91	Poor	Below Average
TMQ	＜1	4	64	74	60--68	68--80	Very Poor	Poor

Note. BI: Before Intervention, AI: After Intervention.

Considerable improvement was also demonstrated on the Functional Skills Scales (FSS) scores of the PEDI after intervention; from 1 to 19 for self-care, from 1 to 12 for mobility, and from 2 to 24 for social functions (Fig. 7). The Care Assistance Scale (CAS) results of PEDI demonstrated that, before the intervention, the infant was completely dependent on the support of the family for functioning, while this caregiver dependence decreased substantially after the intervention (Fig. 8).

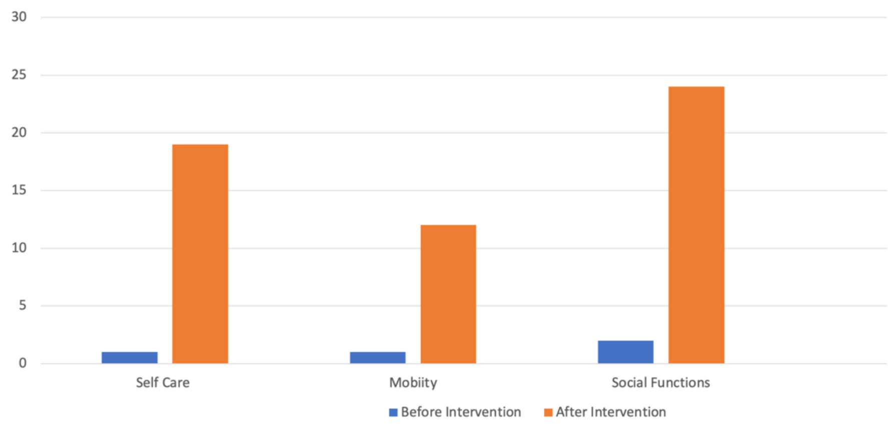

Figure 7. Functional Skills Scales scores of the PEDI before and after intervention.

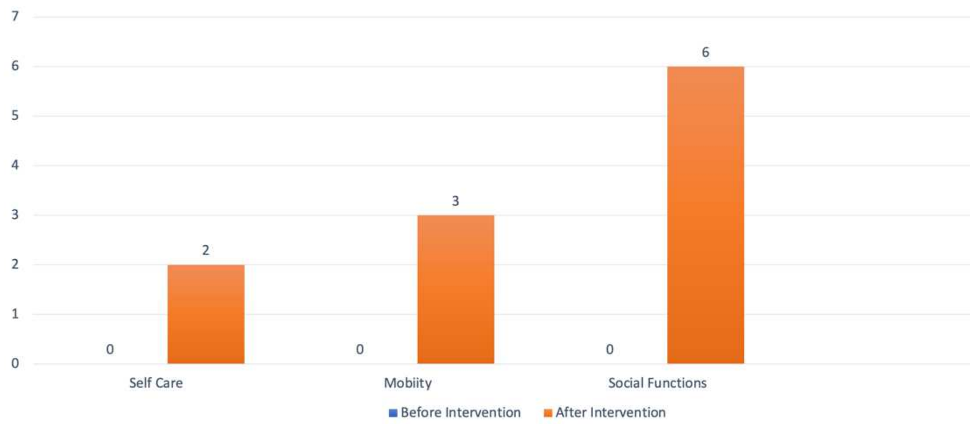

Figure 8. Caregiver Assistance Scales of the PEDI before and after intervention.

BAI scores revealed low anxiety scores for both the mother (13/63 points) and father (14/63) before intervention. These scores did not change after intervention.

Discussion

The effects of EE-based interventions on many neurological conditions have been well demonstrated by experimental studies (Nithianantharajah and Hannan, 2006; Rojas et al., 2013; Marques et al., 2014; Schuch et al., 2016; Griva et al., 2017; Durán-Carabali et al., 2018; 2019b; Jain et al., 2013; Forbes et al., 2020; Laviola et al., 2008). In addition, although all principles do not fully comply with EE, recently developed intervention models inspired

by EE in different neurological populations are promising (Novak et al., 2020; Janssen et al., 2014; Rosbergen e al., 2017; Morgan et el., 2016; Dusing et al., 2018).
This case report is the first attempt to describe a successful application of the HEP® approach as an intervention for an infant with CP. The HEP® approach, a new model based on core principles of EE and ecological theories of development, was presented and preliminary effectiveness of the HEP® approach intervention on regulation, sensory processing, motor development and functional skills in an infant with CP was explored. Results of the 12-month intervention found that the parent's anxiety level remained low; the infant was more regulated and engaged in relationships; the child's sensory sensitivity decreased, gross and fine motor functions developed and all functional skills on the PEDI were improved.

A considerable amount of research clearly reports that the care obligations of a child with a disability may adversely affect caregiver's physical and mental health (Davis et al., 2010). It is also demonstrated that prevalence of anxiety in parents of children with CP is higher than in parents of typically developing children (Barreto et al., 2020). Consideration of parents' mental health is important for developmental goals of children with CP (Gugała et al., 2019), since parent mental health status will effect infant development (Gugała et al., 2019; Grace et al., 2003; Brummelte and Galea, 2016; Oyetunji and Chandra, 2020). Because the HEP approach is an ecological model, parent anxiety level is recognized as a major environmental factor that may affect infant development. The anxiety level of the parents in this case was low before the intervention and this situation did not change after the intervention. A low level of anxiety in parents is a positive support for development and it can be presumed that the HEP® approach intervention does not negatively affect the stress level of the parents. This result may be related to the nature of the HEP® approach which can be considered as a family-centered ecological model that recognizes the importance of the family's well-being to the child's well-being. Family-centered models suggest that every family is unique, is constant in the child's life, and that they are experts in the child's needs (King et al., 2004). Studies have demonstrated that parents who join in programs which are provided in a family-centered way, experience better psychological health, as demonstrated by reduced anxiety, less depression, and higher levels of well-being (King et al., 2004; Lewis et al., 1991; Ireys et al., 2001).

Improved regulatory capacity and engagement of the infant were other outcomes of this study. Homeostasis, which includes regulation, is the first area addressed in the HEP® approach, and many aspects of the intervention principles such as safety, challenge, enjoyment, social and active exploration have ingredients that support self-regulation and engagement of the infant. For these principles to be applied appropriately, the well-being of the parents who provide these opportunities to the infant are central to the intervention. Also, well-being of the parents provides a foundation for positive parenting and child well-being that supports homeostasis (DeGangi, 2000; 2017; Greenspan et al., 2001; Hughes and Baylin 2012; Newland, 2015). Therefore, it can be argued that the improvements in self-regulation and engagement of the infant are at least, in part, a product of successful co-regulatory strategies provided by parents for safety perception, coping with challenges, enjoyment and active expoloration.

Although, not directly addressed in treatment it was found that sensory sensitivity was reduced as a result of intervention. This outcome was not surprising, as dynamic systems theory suggests that small changes in any subsystem (sensory, motor, cardiovascular, etc.) can cause dramatic changes in behaviors not directly related to the initial problem (Thelen and Smith, 1994; Piek, 2006). Accordingly, the developed homeostasis and movement capacity of the individual may have supported the child’s overal arousal level and sensory processing capacity by allowing more self-initiated active sensory experience (DeGangi, 2017; Ayres, 2005; Adolph and Hoch, 2019; Swindeman et al., 2015).

The most obvious improvements in this study was in gross motor and fine motor performance. Both GMFM-88 and PDMS-2 scores improved signficantly compared to the first assessment. Moreover, PDMS-2 quotient interval scores revealed a significant improvement in all quotient areas and the post-intervention GMFCS level I also supported this finding.

Results of this case report are in line with studies which found that EE-based interventions are promising for improving gross motor and manual skills in individuals with CP (Grace et al., 2003; Morgan et al. 2016; Morgan et al. 2013). Morgan et al. (2016) found that “GAME” (Goals - Activity - Motor Enrichment), a motor learning, environmental enrichment intervention resulted in advanced motor and cognitive outcomes when compared with standard care. Novak et al. (2020) in their systematic review suggested that environmental enrichment is an effective intervention to promote task performance in children with CP. Moreover, they stated that high-intensity, fun, motivating, successful, and spontaneous active movements compatible with real-life experiences for meaningful and purposeful goals set by the child or family are key features of successful intervention for children with CP (Novak et al., 2020). The intervention principles of the HEP® approach are fully in line with these key features and may explain the salient improvement in motor performance after HEP® intervention.

It is not surprising then that, enhanced regulatory capacities, sensory processing and motor functions were found in conjuction with improved functional skills. There are many studies that found change in homeostatic, sensory, and motor performance domains affected functional abilities (Durán-Carabali et al., 2021; Bundy et al., 2007; Dunn et al., 2016; Ostensjo et al., 2004; Holsbeeke et al., 2009; Smits et al., 2010). In this case report, performance areas developed with the HEP® approach intervention were found with improvement in the infant's functional skills in self-care, mobility and social function in conjunction with decreased parental support for these activities.

Conclusion

The definition and detailed description of the HEP® intervention, an ecological EE-based approach, was presented for the first time. This case report demonstrated preliminary evidence for the effectiveness of the HEP® approach on self-regulation, sensory processing, gross and fine motor development, functional skills, and caregiver assistance in an infant with CP. Although this case provides information that can be useful for clinicians to understand the nature of providing EE for human babies, because it is a case report, results cannot yet be generalized to the larger population of children with CP. Additional studies are needed to validate the findings

Acknowledgments

The author would like to thank Teresa May-Benson, ScD, OTR/L, FAOTA for valuable feedback and notes on the article.

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