Enriched Environments and Motor Outcomes in Cerebral Palsy: Systematic Review and Meta-analysis

BACKGROUND AND OBJECTIVES: Neuroplasticity evidence from animals favors an early enriched environment for promoting optimal brain injury recovery. In infants, systematic reviews show environmental enrichment (EE) improves cognitive outcomes but the effect on motor skills is less understood. The objective of this review was to appraise the effectiveness evidence about EE for improving the motor outcomes of infants at high risk of cerebral palsy (CP).

METHODS: A systematic review was conducted. Cochrane Central Register of Controlled Trials (PubMed), Cumulative Index to Nursing and Allied Health Literature, Education Resource Information Center, SocINDEX, and PsycINFO databases were searched for literature meeting inclusion criteria: randomized controlled trials; high risk of diagnosis of CP; at least 25% participants aged 2 years; parent or infant interventions post-discharge; and motor outcomes reported. Data were extracted using the Cochrane protocol regarding participants, intervention characteristics, and outcomes. Methodological quality was assessed using risk of bias assessment and GRADE.

RESULTS: A total of 226 studies were identified. After removing duplicates and unrelated studies, 16 full-text articles were reviewed, of which 7 studies met inclusion criteria. The risk of bias varied between studies with the more recent studies demonstrating the lowest risk. Enrichment interventions varied in type and focus, making comparisons difficult. A meta-analysis was conducted of studies that compared enrichment to standard care (n = 5), and totaled 150 infants. A small positive effect for enrichment was found; standardized mean difference 0.39 (95% confidence interval 0.05–0.72; I² = 3%; P = .02).

CONCLUSIONS: EE looks promising for CP, and therefore high-quality studies with well-defined EE strategies are urgently required.

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KEY WORDS: cerebral palsy, infants, enrichment, motor learning

ABBREVIATIONS

CIMT—constraint-induced movement therapy
COPCA—Coping with and Caring for Infants with Special Needs
CP—cerebral palsy
EE—environmental enrichment
GMFCS—Gross Motor Function Classification System
NDT—neurodevelopmental therapy
PDI—Psychomotor Developmental Index
PEDI—Pediatric Evaluation Disability Inventory
PVL—periventricular leukomalacia
QUEST—Quality of Upper Extremity Skills Test
RCT—randomized controlled trial

Ms Morgan and Dr Novak were involved in quality assessment, data extraction, data analysis, and manuscript preparation; Dr Badawi was involved in quality assessment and manuscript critique. All authors were involved in study conception and design, interpretation of data, critical revision, and final approval of the submitted manuscript.

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Children with cerebral palsy (CP) reach ∼90% of their gross motor potential by age 5 and even younger for the more severely impaired, so effective interventions for optimizing early motor development are vital. As with typically developing children, the first 2 years are critically important for cognitive and motor development because the brain is experiencing continuous spontaneous plasticity. Pediatricians, who are responsible for making the diagnosis of CP and referral to rehabilitation, therefore require up-to-date evidence about effective early interventions for children with CP.

The value of enriched environments in enhancing brain recovery at both structural and chemical levels has been repeatedly demonstrated in animal studies. Effects of enrichment include improved memory and motor function. Replication of animal data findings within humans is still undergoing experimentation, with one of the early challenges being how to define an “enriched human environment.” No single agreed definition of environmental enrichment (EE) in human infants exists. In animal studies, an EE is defined as an environment that facilitates enhanced cognitive, motor, and sensory stimulation. Although there are no agreed parameters for enrichment, these animal housing conditions typically include high levels of complexity and variability within their daily lives. In addition, unlike animals, human infants cannot voluntarily access their environment because motor maturation occurs later; for example, ambulation is not present at birth. Consequently infants are dependent on their parents for access to both generalized and motor-specific EE. Much more is known about the negative impact of deprivation on child development, inferring that EE and activity-dependent plasticity are vital.

Animal EE ideas are difficult to replicate within human experiments because humans experience an individualized level of complexity and variability within their daily lives. In addition, unlike animals, human infants cannot voluntarily access their environment because motor maturation occurs later; for example, ambulation is not present at birth. Consequently infants are dependent on their parents for access to both generalized and motor-specific EE. Much more is known about the negative impact of deprivation on child development, inferring that EE and activity-dependent plasticity are vital.

Well-understood examples include the following: (1) institutionalized children within deprived environments display intellectual quotients 20 points lower than peers, which is reversible when EE is applied within orphanages; (2) children living in chronic poverty experience slower growth, worse health, and lower intellectual ability unless EE protective factors are in place (eg, parental responsiveness and acceptance, availability of learning materials, safe play areas, and a variety of experiences); (3) typically developing children experience delayed sitting skills from parents conscientiously following the Sudden Infant Death Syndrome “back to sleep” program, which deprives children of experiences in prone, but is fortunately remediable; and (4) typically developing children experience delayed walking from regular use of infant walkers, whereas Jamaican infants walk earlier owing to parental handling techniques. It should be noted that these latter examples have only a short-term influence on motor development in typically developing children, and it is not known if these environmental influences benefit or disadvantage infants with motor disorders in any way. Motor-enrichment interventions have recently been trialed in preterm and typically developing infants. Reaching training delivered by caregivers to their preterm infants was able to partially ameliorate the delayed reaching skills often observed in the preterm population. Similarly, training parents to practice specific motor tasks with typically developing infants accelerated the rate of motor development in both the short and long term.

In the small amount of literature about the benefits of EE for infants at risk for brain injury, we know that premature infants demonstrate neurobehavioral benefits from sensory-specific EE activities, such as massage and music. Developmental care interventions for premature infants have been shown to deliver modest short-term gains, but with some trials showing no benefit at all. Some programs, such as The Newborn Individualized Developmental Care and Assessment Program, a sensory-specific EE and cue-based intervention for high-risk infants, has been shown to positively influence brain function and motor development. Generic EE via early interventions, such as the Head Start program, provide cognitive benefits short-term, especially for infants from lower socioeconomic backgrounds whose risk of...
Similarly, systematic reviews show favorable short-term cognitive benefits from generic EE programs offered to premature infants.26,27 Because only ~8% to 15% of premature infants will go on to have CP,28 it is not clear whether interventions aimed at preterm infants will have clear benefits for infants with CP. In contrast, “traditional” physical and occupational therapy early-intervention approaches, such as neurodevelopmental treatment (NDT), have not been shown to be effective in improving motor outcomes in infants or older children with CP, despite the theoretical possibility of providing sensory-enrichment cues for learning motor skills.29,30

Given that optimization of neuroplasticity is the aim of all rehabilitation, it is important for those who deliver early-intervention services to understand the parental and EE-intervention role that in turn is informed by knowing the important components of EE for infants with brain injury. Indeed, the importance of the role of parents in providing optimal home environments for at-risk infants,31 as well as arranging opportunities for motor training, have been highlighted in recent reviews.32

Because interventionists use the term EE without definitional or procedural precision, it is important to be clear that not all therapy interventions are enriching. In some standard care interventions, manual handling techniques are applied with the child’s role being largely passive. This contravenes animal EE definitions, in which active exploration of complex and variable environments is required. For the purposes of this review and in the absence of an agreed infant EE definition, we proposed an operational definition of infant EE, consistent with the animal literature (Fig 1). Infant EEs are interventions that aim to enrich at least 1 of the motor, cognitive, sensory, or social aspects of the infant’s environment for the purposes of promoting learning. Examples include interventions aiming to enhance parent-infant interaction, educate parents about assisting their child’s skill development, provide opportunities for active motor learning (self-generated motor activity) by adapting the physical and play environment, or provide comprehensive programs aimed at enrichment across a number of domains.

The purpose of this study was to systematically review the evidence for the effectiveness of EE interventions (either generic EE or motor-specific EE; eg, motor training) for infants at very high risk of CP, which explicitly sought to improve motor outcomes.

METHODS

The method used was a systematic review and meta-analysis with reporting according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement.33 A comprehensive search was conducted of the following databases between May and August 2011 and updated in
May 2012: The Cochrane Library, including the Cochrane Database of Systematic Reviews and the Cochrane Central Register of Controlled Trials (inclusive of PubMed); the Cumulative Index to Nursing and Allied Health Literature; PsycINFO; Education Resource Information Center; and SociNDEX.

Types of Studies

Included studies were randomized controlled trials (RCTs). Systematic reviews were also retrieved but not appraised as per conventions. Controlled studies and studies in languages other than English were excluded.

Types of Participants

Participants were either infants with a confirmed diagnosis of CP of any subtype or severity, or infants designated “at high risk” of CP using the best predictive tools available, namely, abnormal general movements (absent fidgety) or abnormal brain imaging (cranial ultrasound or magnetic resonance imaging). Studies were included in which 25% of participants were 2 years of age or younger at the time of study enrollment.

Types of Interventions

EE Interventions

EE interventions of interest were those in which the infant’s environment was enriched via parent training or coaching in parent-infant interaction or in various stimulation activities, specifically those for motor learning, or where the physical environment was modified, adapted, or constructed to enable motor skill attainment, or where therapists provided intense, targeted motor skill practice aimed at enhancing plasticity. Interventions that solely focused on enriching parent well-being for improving child motor outcomes were excluded. The effects of regular parental caregiving were not specifically teased out for either the EE or comparison group, because if effective for promoting motor development, we would expect as a function of randomization that positive parental caregiving was evenly distributed between both groups. Studies of “NDT plus” were included in the EE categorization if, and only if, the added elements of the intervention (ie, the “plus” component) clearly involved EE.

Comparison Interventions

Comparison interventions were those deemed “standard care” as provided by physiotherapists and included traditional approaches, such as NDT or Vojta. NDT and Vojta were not considered enrichment interventions by our definition, because NDT and Vojta, despite modernization, continue to fundamentally focus on passive therapist-delivered facilitation and inhibition (therapeutic handling).34 In contrast, EE approaches deliberately minimize handling to promote active child-generated muscle activation and movement. Interventions that included handling or positioning embedding into daily routines were regarded as largely passive interventions and were thus treated as non-EE interventions from standard care, on the basis that these treatment ideas originated from NDT.

Types of Outcome Measures

Outcome measures of interest were those that assessed progress in motor skill acquisition at any time point after intervention and as either a primary or secondary measure. To improve homogeneity, meta-analysis was conducted using only data collected at time points immediately at the end of the intervention period.

Search Terms

The following Population, Intervention, Comparison, Outcome search terms were used: P = cerebral palsy OR neonatal stroke OR intraventricular hemorrhage OR brain injury OR hypoxic ischemic encephalopathy OR newborn encephalopathy OR periventricular leukomalacia (PVL); I = environment OR enriched environment OR enhanced environment OR home environment OR nurture OR parenting OR parent training OR parent coaching OR caregiving OR shaping OR stimulation OR intensive task practice; C = nil; O = motor development OR motor skills OR motor learning OR motor outcome. Filters applied during the searching were “infants 0–23 months” and articles in English. Abstracts from conferences and dissertations were excluded. Additional hand searching was conducted from reference lists and field expert recommendations.

Selection of Studies

Two authors (C.M. and I.N.) independently screened all titles and abstracts, identified articles, and excluded irrelevant citations. Full-text articles of all potentially relevant articles were obtained and assessed for eligibility. Ninety-five percent agreement was reached; disagreement was resolved through discussion and consensus. The criteria for study exclusion are documented in Fig 2.

Data Extraction and Management

A data extraction tool based on the Cochrane guidelines35 was used by 2 authors (C.M. and I.N.). The following data were extracted: study design; inclusion and exclusion criteria; participant characteristics, including the diagnosis of CP or “high risk of CP”; number of participants; age and gender of participants; characteristics of the intervention and comparison interventions, including treatment approaches and duration, frequency, and intensity of intervention; details of cointerventions plus compliance with treatment protocol; motor outcomes; methods used to measure change in
motor function; mean scores and SDs of outcomes; and direction of effect for motor outcome. We contacted authors of included studies when there was incomplete reporting of data. All authors contacted were able to provide the missing data requested.

Quality of Studies and Risk of Bias
The methodological quality of the included studies was assessed by using the Cochrane risk of bias recommendations from the Cochrane Handbook for Systematic Reviews of Interventions and is summarized in Table 2.

Analyses
Meta-analysis was conducted for the studies that were clinically homogeneous. Data were analyzed by using Review Manager 5 (RevMan; Computer program Version 5.1. Copenhagen: The Nordic Cochrane Centre, The Cochrane Collaboration, 2012). The I² statistic was used to quantify the heterogeneity of outcomes and informed decisions about whether to pool data. Meta-analyses were conducted by using a random-effects model to conservatively account for the data heterogeneity. The mean differences in motor outcomes were pooled for each study to provide a summary estimate of the effectiveness of EE interventions. For all continuous outcomes with different units, effects were expressed as standardized mean differences and 95% confidence intervals.

RESULTS
The electronic searches, citation tracking, and reference list searches elicited 226 references after 9 duplicates were removed. After screening titles and abstracts, 16 studies were identified, and after inspecting the full-text articles, 7 studies met full inclusion criteria. Reasons for exclusion are summarized in Fig 2.

Included Studies
Across the 7 included studies there were a total of 328 participants (Table 1). Three studies investigated the effects of EE interventions (as per our definition) on very young hospitalized infants with brain injuries and at high risk of CP and followed their progress post discharge. The remaining 4 studies investigated EE interventions (as per our definition) in children older than 1 year with a confirmed diagnosis of CP. The features of EE interventions varied considerably among the studies. Six studies provided part of the EE intervention via parent training or coaching. This included ways of interacting with their infant, strategies for modifying the physical environment for motor task practice, and providing frequent opportunities for task practice. Only 1 study did not actively train parents but encouraged them to “use newly acquired skills when the therapist was not present.”

Methodological Quality and Risk of Bias in Included Studies
The methodological quality and risk of bias in the 7 included studies were variable, with a tendency for the more recent studies to be of the highest quality and at the lowest risk of bias (Table 2). Three studies used adequate methods for generating the randomization sequence but 4 were unclear. Three studies used adequate methods for generating the randomization sequence but 4 were unclear. Three studies used adequate methods to conceal allocation. Blinding of participants and therapists was not possible in any of the studies because of the “hands-on” and thus visible nature of the intervention. Five of the studies used assessors who were blinded to group allocation and 4 studies had
<table>
<thead>
<tr>
<th>Study</th>
<th>Participants</th>
<th>Design and Intervention</th>
<th>Enrichment Used</th>
<th>Provider</th>
<th>Outcome Measures</th>
<th>Results</th>
</tr>
</thead>
<tbody>
<tr>
<td>Badr et al³⁶</td>
<td>n = 62</td>
<td>RCT</td>
<td>Cognitive enrichment: CAMS program</td>
<td>Nurse</td>
<td>Motor: Bayley PDI</td>
<td>No difference between groups, Bayley PDI fell in both groups.</td>
</tr>
<tr>
<td></td>
<td>38M, 24F</td>
<td>Age: 0 mo</td>
<td>Aim: Enhance cognitive and sensorimotor development</td>
<td></td>
<td></td>
<td>Confounded by 30% attrition rate.</td>
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<tr>
<td></td>
<td></td>
<td>Gp 1 = Enriched intensive stimulation using CAMS (Curriculum and Monitoring System)</td>
<td>Motor enrichment: Training parent in various stimulation activities for motor learning</td>
<td>Other: Bayley MDI; NCAFS, NCATS, PSI</td>
<td></td>
<td>NOTE: Data reanalyzed in 2009, showing significant difference between groups favoring enrichment</td>
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<tr>
<td></td>
<td></td>
<td>Gp 2 = Standard care</td>
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<tr>
<td>Law et al³⁹</td>
<td>n = 128</td>
<td>RCT</td>
<td>Motor enrichment: Changing the environment to enable goal attainment</td>
<td>Physiotherapist or occupational therapist</td>
<td>Motor: GMFM, PEDI, joint ROM</td>
<td>No significant difference between groups. Both groups improved equally on the PEDI and GMFM</td>
</tr>
<tr>
<td></td>
<td>79M, 49F</td>
<td>Age: 12-71 mo (34% =2)a</td>
<td>Aim: Improve performance on functional tasks and mobility and increase participation in everyday tasks</td>
<td>Parent coaching in problem solving child movement difficulties</td>
<td>Other: APCP, FES</td>
<td></td>
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<tr>
<td></td>
<td></td>
<td>Gp 1 = context-focused therapy (therapeutic intervention aimed at task ± enriching environment)</td>
<td>Intensive, customized, and variable task practice</td>
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<tr>
<td></td>
<td></td>
<td>Gp 2 = child-focused therapy (therapeutic intervention aimed at child only, may include intensive task practice)</td>
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<tr>
<td>Nelson et al³⁷</td>
<td>n = 37</td>
<td>RCT</td>
<td>Social enrichment: Training parent-infant interaction</td>
<td>Research assistant, then parent</td>
<td>Motor: Bayley PDI</td>
<td>No significant difference between groups on Bayley at 12 mo. Confounded by 25% attrition rate. Best predictor of motor outcome was presence of PVL</td>
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<tr>
<td></td>
<td>18M, 19F</td>
<td>Age: 0 mo</td>
<td>Aim: Reduce incidence of handicap</td>
<td></td>
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<td></td>
<td></td>
<td>Gp 1 = Enrichment via multisensory stimuli + standard care</td>
<td>Sensory enrichment; auditory/tactile/visual/Vestibular</td>
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<td></td>
<td></td>
<td>Gp 2 = Standard care</td>
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<tr>
<td>Ohgi et al³⁸</td>
<td>n = 23</td>
<td>RCT</td>
<td>Social enrichment: Training parent-infant interaction</td>
<td>Neonatal Behavioral Assessment Scale certified examiner</td>
<td>Motor: Bayley PDI</td>
<td>No significant difference between groups on motor outcomes at 6 mo</td>
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<tr>
<td></td>
<td>17M, 6F</td>
<td>Age: 0 mo</td>
<td>Aim: Facilitate infant development</td>
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<td></td>
<td></td>
<td>Gp 1 = Enrichment via training of mother-infant interaction + handling and developmental support using NDT principles</td>
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<tr>
<td></td>
<td></td>
<td>Gp 2 = Standard care</td>
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<tr>
<td>Study</td>
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<td>Results</td>
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<tr>
<td>Palmer et al</td>
<td>n = 48</td>
<td>RCT</td>
<td>Cognitive/social enrichment: Training parent in various stimulation activities</td>
<td>Child development specialist</td>
<td>Motor: Bayley PDI</td>
<td>Significant difference between the groups favoring enrichment at both 6 and 12 mo</td>
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<tr>
<td>(1988)</td>
<td></td>
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<td>Motor enrichment: Changing the environment to enable goal attainment</td>
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<td>Other: Bayley MDI; Vineland</td>
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<td></td>
<td>Age: 12–19 mo</td>
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<td>Gp 1 = Enriched intensive stimulation using <em>learning games</em> (cognitive, sensory, language and motor activities) for 6 mo followed by 6 mo NDT</td>
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<tr>
<td></td>
<td>36M; 12 F</td>
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<td>Gp 2 = Standard care (12 mo NDT)</td>
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<td></td>
<td>100% CP</td>
<td>RCT</td>
<td>Motor enrichment: Intensive, customized and variable task practice</td>
<td>Occupational therapist, physiotherapist or PT assistant</td>
<td>Motor: EBS, PMAL, TAUT</td>
<td>Significant difference between the groups favoring enrichment</td>
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<tr>
<td>Taub et al</td>
<td>n = 18</td>
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<td>Note: Cochrane review in 2007 details QUEST scores on this group not reported in the article</td>
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<td>(2004)</td>
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<td></td>
<td>Age: 7–96 mo</td>
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<td>Gp 1 = Enriched intensive task practice via shaping with constraint</td>
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<td></td>
<td>(50% ≤2)</td>
<td></td>
<td>Gp 2 = Standard care (conventional PT/OT)</td>
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<tr>
<td></td>
<td>13M; 5F</td>
<td>100% CP</td>
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<tr>
<td>Wallen et al</td>
<td>n = 50</td>
<td>RCT</td>
<td>Motor enrichment: Training parent in various stimulation activities for motor learning Intensive, customized task practice</td>
<td>Occupational therapist</td>
<td>Motor: AHA; PMAL-R</td>
<td>No significant difference between groups. Motor outcomes improved equally in both groups</td>
</tr>
<tr>
<td>(2011)</td>
<td></td>
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<td></td>
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<tr>
<td></td>
<td>Age: 19–94 mo</td>
<td>(34% ≤2)</td>
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<tr>
<td></td>
<td>(27M; 23F)</td>
<td>100% CP</td>
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AHA, Assisting Hand Assessment; APCP, Assessment of Preschool Children’s Participation; COPM, Canadian Occupational Performance Measure; EBS, Emerging Behaviors Scale; FES, Family Empowerment Scale; GAS, Goal Attainment Scaling; GMFM, Gross Motor Function Measure; Gp 1, experimental group; Gp 2, control group; LCC, Lack of Confidence in Caregiving; mCIMT, modified Constraint-Induced Movement Therapy; NBAS, Neurobehavioral Assessment Scale; NCATS, Nursing Child Assessment Teaching Scale; PSI, Parenting Stress Index; STAI, State Trait Anxiety Inventory; TAUT, Toddler Arm Use Test.

* Confirmed with author.
adequate follow-up. Only 1 study selectively reported outcomes and 5 studies were free of other bias because they provided full statistical reporting.

Effects of Interventions on Motor Outcomes

Five of 7 included studies compared an EE intervention with standard care and were clinically homogeneous for meta-analysis (ie, compared EE with standard care, and used the Bayley Psychomotor Developmental Index [PDI] [4/5] trials). Standard care was not clearly described in terms of the treatment approaches in use or the intensity of intervention provided. Two of the 7 studies compared 2 different types of EE interventions (as per our definition) head to head.39,42 In the study by Law et al,39 both the context-focused group and the child-focused group enlisted intensive task practice as an EE feature. What differentiated the groups was that the context-focused intervention also included parent training and environmental adaptations to promote functional skill attainment. Likewise, Wallen et al42 compared modified constraint-induced movement therapy (CIMT) with an intensive occupational therapy approach in which both groups received intensive task practice and parent training aimed at EE.

The 5 studies that compared EE with standard care were included in the meta-analysis. Motor outcome data were pooled from 4 studies using the Bayley PDI43 and 1 study using the Quality of Upper Extremity Skills Test (QUEST).44 For the study by Nelson et al,37 only the values reported on infants with a central nervous system injury were included within the meta-analyses, which was possible because these figures were reported separately from infants without central nervous system injury. Data from the 6-month point were used from the Palmer et al40 study because infants in the experimental group received the enrichment intervention only during the first 6 months and then after this they were prescribed maintenance NDT for the next 6 months. QUEST values for the Taub et al41 trial were used, as this was the only motor outcome measure used in this trial for which appropriate psychometrics were available. These values were retrieved from the Cochrane Review by Hoare et al.45 When combined, the 5 studies included a total of 150 participants. The standard mean difference was 0.39 (95% confidence interval 0.05–0.72; I² = 3%; P = .02), indicating a small positive effect favoring enrichment over standard care (Fig 3: forest plot).

**DISCUSSION**

The aim of this systematic review was to determine the effect of EE intervention programs on the motor outcomes of infants who were 2 years and younger with a high risk or diagnosis of CP, compared with standard care. This is the first systematic review and meta-analysis that has attempted to define and measure the effect of EE on motor development of infants with CP. Previous systematic reviews have focused more broadly on motor and cognitive outcomes in preterm populations27 or those at risk for a broader range of developmental disorders.46,47 In these previous studies, favorable cognitive outcomes programs have been consistently demonstrated for a range of early-intervention programs, but motor outcomes rarely improve. Five studies with sufficient homogeneity for meta-analysis were found, which indicated good-quality evidence for a very small but favorable benefit from enrichment interventions in improving motor outcomes for infants with CP. The studies were all RCTs (ie, high levels of evidence, of medium-high quality, and varying levels of risk of bias). The entire body of evidence for EE improving motor outcomes in infants with CP was graded as moderate quality (ie, further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate).48 Nevertheless, our study makes a new and unique contribution to

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**TABLE 2: Cochrane Risk of Bias Assessment**

<table>
<thead>
<tr>
<th>Study</th>
<th>1</th>
<th>2</th>
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<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
</tr>
</thead>
<tbody>
<tr>
<td>Law et al49 (2011)</td>
<td>+</td>
<td>+</td>
<td>—</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>Ohgi et al45 (2004)</td>
<td>+</td>
<td>—</td>
<td>—</td>
<td>+</td>
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<td>+</td>
<td>+</td>
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<tr>
<td>Wallen et al47 (2011)</td>
<td>+</td>
<td>+</td>
<td>—</td>
<td>+</td>
<td>+</td>
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<td>+</td>
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</tbody>
</table>

The Cochrane Risk of Bias assessment domains addressed are the following: (1) random sequence generation; (2) allocation concealment; (3) blinding of participants and personnel; (4) blinding of outcome assessment; (5) incomplete outcome data; (6) selective reporting; (7) other bias. +, low risk of bias; —, high risk of bias; ?, unclear.

**FIGURE 3**: Forest plot.
the literature by highlighting ways to improve motor outcomes at an early age. Unfortunately, it was not possible to draw conclusions about the contributions of the varying components of EE because of the high levels of intervention and participant heterogeneity among the studies. The studies varied in severity of motor impairment, initial degree of risk for CP, the type of EE used, the intervention duration, the involvement of parents, and the motor assessments used. The meta-analysis appears, however, to indicate that enrichment is beneficial, despite differences in child attributes and “favorable” parent characteristics, as conceivably any differences would have been distributed evenly across EE and non-EE groups owing to randomization.

The 2 studies that commenced with newborns were unable to demonstrate favorable motor outcomes for the experimental groups. Although reasons were explored in each publication, we also concluded that these studies were inadvertently underpowered because only a small proportion of participants ultimately ended up with a CP diagnosis. In other words, because most of the participants in both groups were healthy or mildly affected, intervention would be unlikely to affect their results. Infants who have normal or milder motor impairments will unmistakably score better on norm-referenced tools, such as the Bayley PDI, than will infants with CP. Potentially grouping motor-impaired infants with those whose delay is simply related to prematurity does not allow identification of aspects of the interventions that may have been effective for the different diagnoses. In addition, it has been suggested in earlier reviews that norm-referenced tools, such as the Bayley PDI, may not be sensitive enough to measure change in infants with CP.66 The 3 infant studies used different inclusion criteria for defining risk of CP, which is likely to further explain the nil findings. For example, Badr et al66 curiously excluded a subgroup of infants with the highest risk for CP (e.g., Grade IV intraventricular hemorrhage with PVL), but the remaining study group still had some risk factors for CP.

Not surprisingly, only a small percentage of infants were then diagnosed with CP at follow-up. Nelson et al57 reported a final CP diagnostic rate of 44% to 67%, dissolving the study power. None of the studies included infants younger than 12 months who had not been sick in the neonatal period. This is an interesting finding, and supports the authors’ experience that almost half of infants at risk for CP are not being referred for therapy services until closer to their first birthday.29 Another identified limitation in 2 of the infant studies (Nelson et al,57 Oghi et al49), was that the authors ceased intervention before, or at 6 months of age, before the average age at which CP is commonly diagnosed. It is therefore unclear whether ongoing intervention of different types (i.e., EE versus no EE) would have changed the results, as the complexity of motor demands increases over time and children with CP tend to fall farther and farther behind.

Several studies had to be excluded from this review, because they did not meet the inclusion criteria of a sample at high risk of CP; most notably, an RCT that compared the parent coaching intervention “Coping with and Caring for Infants with Special Needs” (COPCA), with standard care. In the COPCA study, there were no differences between the groups with respect to motor outcomes, which should have perhaps been expected given that <25% of participants were eventually diagnosed with CP. Thus, for the most part, authors were comparing healthy infants with healthy infants. Post hoc analysis of infants with CP revealed a positive correlation between PEDIS scores and elements of the COPCA approach.49,50

In the subsequent years since many of these clinical trials have been conducted, the field has learned a great deal more about how to precisely identify infants who are most at risk for CP. It is now possible to identify those infants at risk for CP with a high degree of accuracy using the General Movements Assessment plus imaging.51,52 Abnormal general movements (“absent fidgety”) at 3 months corrected age predicts CP with a sensitivity of ≥92% (specificity ≥82%).53 In light of our study findings, using best practice tools to identify those infants at risk for CP and to tease them apart from those at risk for general delay is very important, as EE interventions can be specifically targeted at motor development if this is expected to be impaired. Also, earlier intervention instituted at a time of greater brain growth and plasticity is likely to be associated with a stronger beneficial effect.

Of the 4 studies that included infants with a confirmed CP diagnosis, the severity of the motor impairment varied, which is known to be a covariate for explaining study findings.54 Only 1 study (Law et al59) included children from all Gross Motor Function Classification System (GMFCS) levels. The Taub et al41 and Wallen et al42 studies included only children with hemiplegia (usually GMFCS I–II) and Palmer et al40 applied their enrichment intervention to a subgroup of children with diplegic CP. Although the Palmer et al40 study predates the invention of GMFCS, it is clear from the description of the participants that almost all infants had motor skills that fall into GMFCS I to III categories (i.e., were certain to be ambulatory). Broadly speaking, the interventions described in these 4 studies all involved motor task practice customized to the child, delivered by...
a professional (therapist or teacher) and reinforced by tailored home practice. Interestingly, these 4 of the 7 studies were the studies that showed a positive trend favoring EE.

The study by Law et al. that compared 2 different EE interventions head-to-head found both approaches were equally effective. Law et al. findings are consistent with other studies of functional therapy or task-based training EE approaches known to be effective in older children. In line with the International Classification of Functioning, Disability, and Health, functional therapy or task-based training EE approaches deliberately consider the impact of the environmental context in the design and implementation of therapy. The difference with Law et al. context-focused study is that 2 novel approaches are compared: “hands off and hands on.” In a typical clinical situation it is unlikely that only child-focused (“hands on”) or context-focused therapy (“hands off”) would be provided. A combination of strategies that target both the child and the context is more likely. Our review did not locate any studies that used these functional motor learning, goal-driven, and environmentally enriching approaches for infants with little or no motor repertoire. This remains a gap in literature, warranting further study. Wallen et al. and Taub et al. used different models of CIMT as a form of EE (as per our definition). It is the motor-learning strategies, or shaping, that co-occur with use of a constraint that make this approach motor-specific enrichment. The 2 studies used quite different approaches with variations in intensity and the type of constraint used. However, both experimental groups offered a similar total amount of intervention (mean 119 hours) but over different durations (3 weeks or 10 weeks). Although the study by Taub et al. demonstrated impressive motor outcomes for the constraint group, a subsequent Cochrane review outlines substantial sources of bias in this study. In contrast, both groups in the Wallen et al. study used an EE approach in which the experimental group constraint was the “added extra.” Motor outcomes improved in both groups. It may be that the consistent motor-learning/task-practice approach is the key component of these studies.

**Limitations of This Review**

Some of the included studies in this review did not provide adequate descriptions of standard care interventions, resulting in the possibility that enrichment activities were indeed part of these comparison groups, which would ultimately dissolve statistical power. It is, however, our experience that standard care for young infants is typically a “wait-and-see” approach, which mostly involves active monitoring of the infant over the first 12 months. It is also possible that because of the definition of EE used, intervention studies that actually offered enrichment were omitted. This confounder was minimized by clearly defining EE and features of enrichment, using extensive hand searching and using search terms indicative of the early intervention field. In particular, opportunities for motor task practice were included within the definition of EE, as it seems evident that for infants to develop motor skills, opportunities must be provided within their learning environment. However, other definitions of EE may single out EE from task practice opportunities. Future studies should therefore be careful to detail the approaches and strategies in use, the frequency and intensity of intervention of all groups, and account for the effect of cointerventions. In particular, the breakdown of the approach and the extent of parent involvement should be specified to advance our understanding of human EE.

**CONCLUSIONS**

Enrichment interventions to improve motor outcomes in infants at high risk of CP appear promising. Therefore, more high-quality, low-bias, large-sample, longitudinal RCTs that examine the effects of motor task practice with deliberate attention to environmental enrichment via appropriate parent training and a variety of stimulating opportunities for learning are urgently needed. Researchers also need to use the best available evidence to accurately identify those at the highest risk of CP for inclusion in these trials to ensure adequate study power.

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Enriched Environments and Motor Outcomes in Cerebral Palsy: Systematic Review and Meta-analysis
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