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Movement-based interventions for preschool-age children with, or at risk of, motor impairment: a systematic review

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ABBREVIATIONS

CIMT	Constraint-induced movement therapy
DCD	Developmental coordination disorder
ICF-CY	International Classification of Functioning, Disability and Health: Children & Youth version
PEDro	Physiotherapy Evidence Database
RCT	Randomized controlled trial
SMD	Standardized mean difference

AIM To explore the efficacy of movement-based interventions to improve motor skills in preschool-age children with, or at risk of, motor impairment, including those with a diagnosis of cerebral palsy, autism spectrum disorder, and developmental coordination disorder.
METHOD Relevant electronic databases were searched for randomized or quasi-randomized controlled trials. Outcomes were classified using domains of the International Classification of Functioning, Disability and Health: Children & Youth version. Quality was assessed using the Physiotherapy Evidence Database scale. Risk of bias was assessed using the Cochrane Risk of Bias tool. Effect sizes were calculated using Cohen's *d*.
RESULTS Seventeen articles exploring a heterogeneity of intervention types, population groups, and outcome measures met the inclusion criteria. Movement-based interventions did not significantly improve outcomes in either the body structure and function or activity domains in most studies. No studies used a participation outcome measure.
INTERPRETATION There is a paucity of evidence exploring movement-based interventions in the preschool-age group. Although movement-based interventions showed potential for improving body structure and function and activity outcomes for children with motor impairment, results were mostly not significant. Small sample sizes, variable study quality, and risk of bias limit confidence in the results.

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Review

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What this paper adds

- The evidence is inconclusive to support movement-based interventions in this group.
- No studies used outcome measures assessing participation.
- Variability in intervention type and study quality limit confidence in results.

[Main text]

Preschool age (3–6y) is a critical period for the development of motor skills[1] and an opportune time to deliver motor interventions.[2] Targeting intervention before school entry may improve school readiness. Delay or impairment in motor skill acquisition can negatively affect a child's ability to participate in activities of daily living,[3] and is associated with impaired academic, cognitive, and socio-emotional outcomes.[4,5] Preschool-age children with motor impairment participate less frequently than typically developing children in play[6,7] and physical activity.[8] Importantly, poor motor skill competence during childhood has been associated with lower levels of physical activity throughout adolescence and into adulthood.[9] For children with, or at risk of, motor impairment, effective interventions are needed to support the development of motor skills. Movement-based interventions, in which children actively participate in therapy, may facilitate improvements in motor skills and provide opportunity to learn, practise, and consolidate these skills.[2,10]

Previous systematic reviews for children with motor impairment have focused on specific diagnoses, such as cerebral palsy (CP)[11,12] or developmental coordination disorder (DCD),[10,13] or focused on specific populations, such as children born preterm,[14] who are at risk of motor impairment.[15] While some interventions are diagnosis specific, others, including movement-based interventions aiming to improve motor skills, may have capacity to improve outcomes for children with a range of diagnoses. Limiting a review by diagnosis or population type risks missing those interventions that can be applied to a broader group of children. It fails to recognize that, when choosing an intervention, we do not choose on the basis of the diagnostic label (e.g. DCD) but rather on the functional disability, or the interaction between the health condition and the context (e.g. balance difficulties riding a bicycle).[16,17] A focus on functional outcomes is supported by findings that a child's functional characteristics, such as their ability to perform motor skills, better explain a child's support needs than diagnosis.[18,19] There is therefore a compelling argument for considering interventions from the perspective of the desired outcome, to improve motor function, rather than the diagnosis of the child. This is particularly important for the preschool age group, for which there are limited systematic reviews to date. Previous reviews have focused on early intervention during infancy[14,20] or, if considering older children, have often included participants across a broad age range,[10,11,13,21] which poses a limitation when translating the findings to children of preschool age. One previous systematic review has specifically considered motor skill interventions for preschool-age children, but included only children with developmental delay.[22] To our knowledge, there are no systematic reviews looking at movement-based interventions for children with, or at risk of, motor impairment that specifically focus on the preschool age group. Considering how important the preschool period is for motor development,[1] this represents a significant gap in the literature.

This systematic review examined the efficacy of movement-based interventions to improve motor skills in preschool-age children with, or at risk of, motor impairment. We aim to present the effects of interventions systematically, by classifying outcomes using the International Classification of Functioning, Disability and Health: Children & Youth version (ICF-CY) domains of body structure and function, activity, and participation.[23] Classification using the ICF-CY domains enables the effect of an intervention to be described in a way that is meaningful and widely comprehended by healthcare practitioners, facilitating a better understanding of the constructs improved through therapy.

METHOD

This systematic review was undertaken according to PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines.[24] The protocol for this systematic review was registered with the PROSPERO International Prospective Register of Systematic Reviews before starting (CRD42018090141).

Search strategy

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The search strategy was developed and reviewed by all authors. Electronic searches were performed (by KLC) in relevant databases including MEDLINE, CINAHL, Embase, Cochrane Collaboration, PsychInfo and Physiotherapy Evidence Database (PEDro) up to July 2018. Reference lists of included studies were scanned for further eligible studies. We used discrete search strategies for each database with terms relevant to motor impairment, diagnoses and movement-based interventions. The full search strategy is provided in Appendix S1 (online supporting information).

Eligibility criteria

Included studies fulfilled the following criteria: (1) randomized or quasi-randomized controlled trial (RCT); (2) participant mean age and standard deviation between 3 and 6 completed years; (3) children with motor impairment, including those with a diagnosis of CP, autism spectrum disorder, Down syndrome, DCD, developmental delay, or fetal alcohol spectrum disorder; children born preterm (<37wks gestation) or low birthweight (<1500g); or children with motor impairment (below the 16th centile) according to a standardized assessment tool; (4) the primary aim of the intervention was the improvement of motor function, and the principal component was movement that was actively participated in by the child; (5) comparison interventions were movement-based interventions, standard care, or no intervention; (6) one or more primary outcome assessed motor function.

Studies were excluded if the intervention or comparison included drug therapy or surgery, or if the primary aim was not the improvement of motor function. Studies that included therapies, such as passive stretching, in which the child was not actively engaged, or studies exploring the effect of a non-movement-based component of the therapy, such as an assistive device, were excluded. Studies not published in English were excluded.

Inclusion and exclusion criteria were applied independently by two authors (KLC and RAA). The titles and abstracts of all articles elicited by the search strategy were screened for eligibility. Articles that were not excluded at this stage then underwent full text review. Disagreements during the screening and full text review were resolved by a third, independent reviewer (AJS). Study authors were contacted for any missing information.

Data extraction and ICF-CY coding

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Data were extracted by two independent reviewers (KLC and RAA). We extracted information on the trial design using items based on the template for intervention description and replication (TIDieR) checklist, including type of intervention, duration, frequency, dosage, therapy provider, and location.[25] Participant characteristics, outcome measures, study results, and adverse events were also extracted. For studies that reported outcomes at multiple time-points, or used cross-over or waitlist designs, results were taken from the first assessment after intervention.

Motor outcomes were categorized using the ICF-CY coding rules proposed by Cieza et al.[26] and Fayed et al.[27] First, each item in the outcome measure was coded to a domain: (1) body structures (s), (2) body functions (b), or (3) activity and participation (d). The ICF-CY does not distinguish between the activity and participation domains; both are coded (d).[23] Therefore, to delineate these domains, we applied the definition of participation established by Coster and Khetani to each item coded (d).[28] This definition uses the ICF-CY definition of participation, 'involvement in a life situation';[23] however, it expands upon 'life situation' to mean 'sets of organized sequences of activities directed toward a personally or socially meaningful goal'.[28] This method has previously been used to code outcome measures for children.[29,30] Finally, outcome measures were classified as the domain that was coded most frequently within each measure.

Quality assessment and risk of bias

The methodological quality of included studies was assessed using the PEDro scale,[31] while risk of bias was assessed using the Cochrane Risk of Bias Tool.[32] Two independent assessors (KLC and RAA) applied both tools to each included study. Studies with confirmed PEDro scores according to the PEDro were allocated that score.

Narrative synthesis and data analysis

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Intervention design was reported through narrative synthesis using the TIDieR checklist.[25] Study results were presented in ICF-CY domains as determined by outcome measure. Owing to the heterogeneity of intervention types and outcome measures, a meta-analysis was not appropriate. Instead, where possible, continuous data for each study were presented using standardized mean difference (SMD) and 95% confidence intervals (CIs).[33] SMDs were calculated from the mean and standard deviation of each group from the first time-point after intervention using Cohen's *d*.[33] If the mean and SD of outcome were not available (and authors were not able to provide additional data), results were presented as reported in the original article.

RESULTS

The search strategy yielded 988 papers, of which 123 were considered for full text review. A total of 17 papers fulfilled inclusion criteria, comprising 14 studies. In three cases, multiple articles discussed outcomes from a single RCT,[34–40] while one article[41] contained results from two separate studies. Study selection is outlined in a PRISMA diagram (Figure S1, online supporting information). Study characteristics of the included articles are provided in Table S1 (online supporting information).

Quality assessment and risk of bias

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Overall study quality and risk of bias was variable (Table S2, online supporting information). PEDro scores ranged from 3[39,42] to 8 out of a possible 10.[43,44] Seven papers did not adequately blind outcome assessors,[36,40–42,45–47] three papers had inadequate follow-up,[42,47,48] between-group differences were found in three articles at baseline,[38,39,41] and only five studies fulfilled intention-to-treat criteria.[36,38,43,44,46] Key concerns for risk of bias included high risk of bias for randomization (four articles),[39,40,42,46] and high[39,40,42,46,49] or unclear[34–36,41,45,47,48] allocation concealment for most studies. Two studies had high risk of detection bias,[45,46] attrition bias was high[41,48] or unclear[42,47] in four studies, while two studies did not report results for all outcomes.[41,49] As is common in movement-based therapies, no studies were able to blind therapists or participants. This lowered PEDro scores and resulted in high risk of performance bias across all included studies. The risk of bias tool was applied for each of the two interventions described within the article by Valentini and Rudisill.[41]

Interventions

Intervention type, duration, dosage, and provider are outlined in Table S1. Motor skills groups were the most common intervention (six studies),[39–41,45,46,48] followed by constraint-induced movement therapy (CIMT).[34,35,37,38,43] The remaining studies explored treadmill training,[49] arm cycling,[42] conductive education (movement-based therapy for children with CP assuming that CP is a learning problem),[44] myofeedback,[47] and balance, strength, and aerobic training using a Nintendo Wii.[50]

Seven studies delivered interventions in a group format,[34–36,39–41,44–46,48] with most implementing a standardized protocol.[39,40,46,48] Two studies exploring motor skills groups indicated that children were encouraged to choose what activity they practised, and for how long, emphasizing the benefits of child-initiated therapy.[41,45] Six studies provided the intervention individually.[37,38,42,43,47,49,50] Most emphasized an individualized programme, with intervention components chosen or adapted to suit each child,[37,38,43,49,50] while two studies delivered a standardized protocol.[42,47] A combined approach was adopted by a study evaluating a CIMT intervention, which comprised exercises performed as a group as well as individual therapy.[34–36]

Overall, interventions were described with variable detail. Two CIMT studies emphasized motor-learning principles and functional activities[34–38] while another indicated that children practised 'designated task movement' but did not expand upon what these movements might be.[43] Motor-learning principles were also emphasized in a Nintendo Wii intervention[50] and a group intervention for children with DCD.[39,40] Descriptions of motor skills groups emphasized fundamental motor skills, with most articles providing examples (e.g. running, throwing a ball).[39–41,45,46,48] Studies considering conductive education[44] and arm cycling[42] were described in less detail; both provided equipment lists, but limited information on what the intervention involved. A study evaluating individualized, facilitated gait training provided no information on amount of bodyweight support or treadmill speed, limiting reproducibility of the study.[49] Compared with interventions, control therapies were less well-described. Several comparison groups were described as having 'conventional therapy',[44] 'gait training',[42] or 'traditional rehabilitation',[50] with little additional information provided. Four studies provided brief details on exercises or activities involved in the control therapy.[34–36,43,47,49] The remaining studies compared different dosages,[37,38] instructional methods,[41,45] no therapy,[41,46] or consultation and advice from a physiotherapist.[39,40]

Body structure and function outcomes

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Body structure and function outcomes were assessed in seven studies. SMDs and 95% CIs are presented in Figure S2 (online supporting information). Studies explored various parameters including gait,[42,49,50] upper limb range of motion,[35,47] upper limb strength,[47,50] and quality of upper limb function.[35,37,43] Most outcomes demonstrated no between-group differences, although there was an overall trend in favour of the intervention group. The exception was for two studies that demonstrated significant differences in outcomes in favour of intervention. Large, significant effect sizes for wrist extension (SMD 1.24, 95% CI 0.46–2.03) and triceps strength (SMD 1.18, 95% CI 0.40–1.95) were found in a study considering myofeedback in addition to exercise and isokinetic training[47] and in another study assessing Nintendo Wii training (grip strength: right, SMD 0.74, 95% CI 0.10–1.39; left, SMD 0.66, 95% CI 0.02–1.30).[50]

Activity outcomes

Twelve articles evaluated interventions using activity outcomes. SMDs and 95% CIs are presented in Figure S3 (online supporting information). Most results showed no significant differences between the intervention and the comparison groups, with a trend favouring the intervention group performing better. Results from a study by Apache[45] are not included in the figure as it was unclear which group received the intervention first in a crossover study (the author could not be contacted to clarify).

Gross motor skills were assessed in eight studies, with all studies finding greater improvements in the intervention group than in the comparison group.[40,41,44,46,48–50] Large effect sizes supported motor skills groups for children with motor impairment in two studies,[41,48] but were not significant in a further two studies (Fig. S3).[40,46] A large, significant effect size was estimated in the Test of Gross Motor Development, locomotor subscale, in a study comparing instructional methods in group motor skills, while a small, non-significant effect size was found for the object control subscale (Fig. S3).[41] Moderate but non-significant effect sizes were estimated for gross motor outcomes in studies assessing treadmill bodyweight support[49] and Nintendo Wii training (Fig. S3).[50]

Upper limb function was assessed in two RCTs.[34–37] Large, significant effect sizes were estimated favouring modified CIMT compared with usual care in two outcome measures.[34,35] The remaining upper limb function outcomes for this study produced small to moderate effect sizes that were non-significant (Fig. S3).[36] In contrast, the effect sizes for an RCT comparing CIMT dosage favoured the larger dosage, but did not reach statistical significance at the post-intervention assessment (Fig. S3).[37]

Goal-based outcomes were assessed in a study comparing modified CIMT with usual care.[34] After intervention, 82% of children in the intervention group but only 23% in the comparison group had a greater than 2-point increase in their Goal Attainment Scale score (indicating that the goal had been reached or exceeded).[34] While the specific goals are unknown, the study authors indicate the goals were functional, and thus they probably fit in the activity domain.[34] Large, significant effect sizes were estimated on both the satisfaction and performance subscales of the Canadian Occupational Performance Measure favouring the intervention group.[34]

In all remaining activity outcomes, there were no significant differences between the intervention and comparison groups, with two exceptions. Large, significant effect sizes were found for the single leg stance test supporting Nintendo Wii training (SMD 0.78, 95% CI 0.14–1.43),[50] and for perceived motor competence in a study comparing instructional methods for group motor skills (Harter and Pike Scale, SMD 0.91, 95% CI 0.25–1.57).[41]

DISCUSSION

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This review used broad inclusion criteria to capture all available evidence exploring the efficacy of movement interventions for preschool-age children with, or at risk of, motor impairment. Only 14 quasi-randomized or RCTs fulfilled the inclusion criteria, comprising seven types of movement-based intervention. Half of the included studies considered interventions for children with CP. Some interventions, such as conductive education or CIMT, are only applicable to specific groups of children, rather than a broader group of children with motor impairment. The heterogeneity of included studies made it difficult to establish any definite comparisons or conclusions. For many studies, we found no significant differences between the intervention and comparison groups, although there was a trend towards the intervention group. While this review does not provide conclusive evidence, it does highlight the paucity of RCTs that explore motor interventions for preschool-age children.

Overall, we found that movement-based interventions did not significantly improve outcomes in the body structure and function domains. In cases where large differences were estimated, careful interpretation taking into account variable risk of bias and study quality is warranted. Results should be considered with caution for a study considering arm cycling in addition to gait training because of low study quality, high risk of selection bias, and no reporting on attrition rates.[42] Large effect sizes were estimated in one study for outcomes favouring myofeedback; however, this study was of poor quality and provided little information on blinding of outcome assessors or attrition rates, possibly biasing results.[47] Clinical significance should also be examined when interpreting results. Although effect sizes were large and significant for grip strength,[50] a between-group difference of less than 1kg is unlikely to be clinically meaningful. Likewise, a large effect size was found for the single leg stance test (activity outcome) after Nintendo Wii training; however, this reflects a between-group difference of 1 second.[50] Minimal clinically important differences, however, are not known for either measure in this population.

Movement-based interventions were found to improve outcomes in the activity domain in fewer than half of outcomes. Large effect sizes were found to support modified CIMT in upper limb function and goal-based outcomes.[34,35] This study[34,35] was of good quality and low risk of bias, and its results reflect previous systematic reviews supporting CIMT for children with CP.[12] Large, significant effect sizes were estimated supporting motor skills groups in three studies,[41,48] but these results should be interpreted cautiously. One study, in particular, was of low quality and high risk of detection, attrition, and reporting bias.[41] Significant between-group differences reported in some baseline measures in this study limited confidence in the effect sizes we calculated.[41]

No included studies used participation outcomes. This may be because authors did not expect interventions to affect participation, or because suitable participation measures were unavailable, rather than because participation measures were actively excluded. However, clinicians are encouraged to consider participation when providing therapy for children with motor impairment;[51] thus the scarcity of interventions assessing participation at preschool age represents a concerning gap in the literature. This was not an unexpected finding; previous systematic reviews have commented that participation outcomes are rarely used for children with motor impairment.[21,52–54] Participation is an important rehabilitation goal, and there is minimal evidence to suggest participation is an inevitable consequence of interventions targeted at the activity or body structure and function levels.[21,55,56] Interventions that target and assess participation are therefore needed for preschool-age children with motor impairment to support therapy providers and families in clinical decision making.

Most included trials reported details on duration, dosage, location, and who delivered the intervention. Minimal insight can be gained into the effect of therapy characteristics on activity outcomes. The longer CIMT programme of 8 weeks[34–36] produced larger effect sizes than the 4-week programme;[37] however, no definite conclusions can be drawn as the latter compared a smaller dosage, rather than a usual-care comparison group. Motor skill groups were all of similar duration, and there did not seem to be a better outcome from 2 hours compared with 1 hour of class. Although largest effect sizes for interventions targeting body structure and function outcomes were found for the two studies with the longest durations (6mo),[42,47] both studies were of low quality, and any associations between duration and effect should thus be considered with caution. The details of the interventions themselves and what they involved were not consistently described. Clear descriptions of interventions are necessary for reliable replication by clinicians and other researchers.[25] Only two included RCTs reported on adverse events, with both studies indicating that none occurred.[37,38,50] While movement-based interventions should not pose a significant risk to children, adverse events should still be reported as they can influence clinical decision making and risk management strategies.

Overall, the results of this review should be interpreted with caution. Small sample sizes across almost all the included articles are likely to have produced results that were inadequately powered, and therefore may have resulted in type 2 errors. In addition, small sample sizes may explain the wide 95% CIs estimated in effect size calculations, leading to imprecise results that reduce the generalizability of findings. Quality and risk of bias of included articles were variable, limiting the robustness of the conclusions resulting from this review. Previous systematic reviews considering task-specific training[13] and motor-based interventions[10] for children with DCD have reported similar findings in regards to small numbers of studies of heterogeneous quality and intervention type, and likewise found difficulties establishing definite conclusions. Movement-based interventions with emerging efficacy for older children with motor impairment, such as task-specific training[52] or goal-directed interventions based on motor-learning principles,[12] require further investigation to determine whether they are effective for children of preschool age. Similarly, early intervention, which improves motor outcomes for infants,[14] may also be effective if implemented with children at preschool age. These interventions may need to be adapted to suit preschool children's motor and cognitive abilities; further good quality research is therefore necessary to establish the efficacy of interventions for preschool-age children with motor impairment.

Strengths and limitations

The ICF-CY framework has been critiqued for not clearly delineating between the activity and participation domains.[28,55] This distinction is imperative as it is necessary to provide a common understanding between clinicians and researchers targeting interventions towards participation.[57] A particular strength of this systematic review is the use of a clear definition of participation which addresses the ambiguity between activity and participation.[28] Another strength is the use of an established ICF-CY coding method.[26] Outcome measures have not typically been developed with the ICF-CY in mind, however, and therefore cannot always be easily fitted into ICF-CY domains. One outcome, a non-standardized questionnaire on physical activity participation, could not be coded as we were not successful in contacting the authors.[41] A limitation of this review was including only articles written in English. In addition, publication bias may have influenced the number of available eligible articles.

CONCLUSION

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This systematic review highlights the limited evidence available exploring interventions to improve motor function across the activity and body structure and function domains of the ICF-CY for preschool-age children with motor impairment. Further research is needed for this important age group to support clinicians in providing evidence-based therapy to children and their families. Of particular concern, there were no studies using participation outcomes. Understanding how therapy can affect participation is important for maximizing opportunities for children with motor impairment to continue to develop their motor skills.[55] High-quality trials with participation-focused outcome measures are needed to support clinicians providing therapy to preschool-age children.

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SUPPORTING INFORMATION

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The following additional material may be found online:

Appendix S1: Search strategy.
Figure S1: PRISMA flow diagram.
Figure S2: Effect of movement-based interventions on body structure and function outcomes.
Figure S3: Effect of movement-based interventions on activity outcomes.
Table S1: Study characteristics of included articles
Table S2: Study quality and risk of bias

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