A Critical View on Motor-based Interventions to Improve Motor Skill Performance in Children With ADHD: A Systematic Review and Meta-analysis

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A critical view on motor-based interventions to improve motor skill performance in children with Attention Deficit Hyperactivity Disorder: A systematic review and meta-analysis

Running head: Motor-based interventions in children with ADHD

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Abstract

Objective: To map the effect of motor-based interventions on motor skills in children with ADHD.

Method: A systematic literature search was performed in Pubmed, Web of Science and the SCOPUS database (last search: October 30th 2022). Methodological quality was assessed using the PEDro-scale and the quality of evidence determined with the GRADE-method. Meta-analysis was performed when at least five studies were available.

Results: Thirteen studies (7 RCTs) satisfied the inclusion criteria, five of which were eligible for meta-analysis. Only one of the included studies reached the low risk of bias threshold. Comparing different motor-based interventions to any non-motor control intervention showed large motor skill improvements (SMD=1.46; 95% CI=[1.00;1.93]; I^2=47.07%). The most effective type of motor-based intervention and the optimal treatment parameters could not be determined yet.

Conclusion: Motor-based interventions in general seem to improve motor skills in children with ADHD. Additional RCTs are needed to increase current low GRADE confidence.

Body of text

1. Introduction

With a worldwide-pooled prevalence of 5.3% (Polanczyk, de Lima, Horta, Biederman, & Rohde, 2007), Attention Deficit Hyperactivity Disorder (ADHD) is one of the most prevalent neurodevelopmental disorders in childhood. ADHD typically manifests early in development and is characterized by developmental deficits that produce impairments of personal, social, academic and/or occupational functioning (APA, 2013). Core features of the disorder are (a combination of) hyperactivity/impulsiveness and inattention (APA, 2013). Children with ADHD also present with slower rates of information processing (Cortese, 2012), resulting in poorer performance on standardized tests and lower grades at school, thereby increasing their chances of early school drop-out (Weibel et al., 2020). Furthermore, 30-60% of the children with ADHD (Fliers, Franke, & Buitelaar, 2011; Gillberg et al., 2004; Goulardins, Marques, & De Oliveira, 2017) have fine and gross motor difficulties, even after their ADHD symptoms are managed (Goulardins et al., 2017). They encounter difficulties with handwriting, ball skills, (bimanual) coordination, balance, gait and motor planning (Goulardins, Marques, Casella, Nascimento, & Oliveira, 2013; Kaiser, Schoemaker, Albaret, & Geuze, 2015; Mao, Kuo, Yang, & Su, 2014; Meyer & Sagvolden, 2006; Papadopoulos, McGinley, Bradshaw, & Rinehart, 2014), in such a way that they perform significantly poorer than their typically developing peers on standardized norm-referenced motor scales (Cho et al., 2014; Jeyanthi et al., 2016, Geuze 2005). Inadequate motor skill acquisition may lead to lower academic achievement, low self-esteem and participation avoidance, causing a vicious circle of inactivity, low physical fitness, sedentary behavior and eventually social isolation (Mazzone et al., 2013; Smits-Engelsman & Verbecque, 2021) or even a tendency for substance abuse during adolescence (Jeyanthi, Arumugam, & Parasher, 2019; Weibel et al., 2020). Inevitably, secondary problems such as overweight and noncommunicable diseases may emerge (Smits-Engelsman & Verbecque, 2021). This negative spiral, that may manifest itself at different levels of functioning, should therefore be broken as early in life as possible.

International clinical practice guidelines for treating children with ADHD recommend both pharmacological and non-pharmacological approaches (Coghill et al., 2021; Wolraich et al., 2019). Medication (i.e. stimulants) has large beneficial short-term effects on the core features of ADHD, thereby improving their overall daily functioning - especially in a school context. The long-term effects, however, are not clear yet (Coghill et al., 2021; Wolraich et al., 2019). Furthermore, many adverse effects are known to stimulant medication, such as appetite suppression, abdominal pain, mood and sleep disorders and headaches (Dalrymple, McKenna Maxwell, Russell, & Duthie, 2020; Wolraich et al., 2019). Children taking higher dosages of medication even seem to suffer from growth retardation (Dalrymple et al., 2020; Wolraich et al., 2019). Since stimulants tend to increase heart rate and blood pressure, there may also be long-term cardiovascular consequences, though not confirmed yet in research (Wolraich et al., 2019). This has led to a growing concern among parents and caregivers, making them explore other, non-pharmacological options (Berger, Dor, Nevo, & Goldzweig, 2008). Such options comprise behavioral parent training, counselling, social skill training, neurofeedback, school-based contingency management, dietary interventions and cognitive training, all for which no strong scientific recommendations are available (Coghill et al., 2021; Dalrymple et al., 2020; Wolraich et al., 2019). Interestingly, no recommendations have been made towards motor-based interventions. The available research merely provides evidence for the beneficial impact of such interventions on the core features of ADHD (Cerrillo-Urbina et al., 2015; Jeyanthi et al., 2019; Ng, Ho, Chan, Yong, & Yeo, 2017; Song, Lauseng, Lee, Nordstrom, & Katch, 2016; Vancampfort, Scheewe, van Damme, & Deenik, 2020; Villa-Gonzalez, Villalba-Heredia, Crespo, del Valle, & Olmedillas, 2020; Xie et al., 2021) and executive functioning (Cerrillo-Urbina et al., 2015; Liang, Li, Wong, Sum, & Sit, 2021; Suarez-Manzano, Ruiz-Ariza, De La Torre-Cruz, & Martínez-López, 2018; Welsch et al., 2021). Despite the fact that some systematic reviews and meta-analyses incorporated motor skills as an outcome for determining the efficacy of motor-based interventions, it was not their primary aim, which results in an incomplete overview of current literature (Neudecker, Mewes, Reimers, & Woll, 2019; Suarez-Manzano et al., 2018; Sun, Yu, & Zhou, 2022; Villa-Gonzalez et al., 2020).

Currently no strong evidence on the effectiveness of motor-based interventions to improve motor skill performance in these children is available, despite the fact that children with ADHD do exhibit motor skill problems, accompanied by adverse secondary problems affecting their overall functioning. As such, for children with ADHD no guidelines on motor-based interventions are available (Shah, Sagar, Somaiya, & Nagpal, 2019; Wolraich et al., 2019). This review aims to fill this knowledge gap by mapping the evidence for available intervention studies addressing motor skill deficits in children with ADHD. First, we aimed to determine the effectiveness of any type of motor-based intervention compared to a non-motor control (e.g. waitlist, no intervention, cognitive tasks or medication). Secondly, we aimed to identify the best choice of specific types of motor-based interventions.

2. Methods

2.1. Protocol and registration

This review (PROSPERO registration number: CRD42022310085) follows the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Page et al., 2021).

2.2. Search

The electronic databases of PubMed, Web of Science and SCOPUS were systematically searched (last search: October 30th, 2022) using controlled terminology and free text terms (Appendix S1). The references of included studies and relevant reviews identified through the search, were screened.

2.3. Study selection

Relevant studies were identified in two screening phases (title/abstract and full text) using predefined selection criteria (order: population, intervention, outcome, study design and language). Two independent reviewers (LK, EV) selected the studies. In case of ambiguity during the first screening phase, studies were screened on full text. Discrepancies regarding screening were discussed until consensus was reached.

The following selection criteria were applied:

1. *Population*: Children between 2 and 18 years old, diagnosed with ADHD. Papers mentioning Developmental Coordination Disorder (DCD) as a comorbidity were excluded as all children with DCD experience motor skill difficulties, thereby potentially distorting the results.
2. *Intervention*: The training was performed in a real-life environment aiming at improving motor skill performance. The performed intervention needed to be clearly described, allowing reproduction of the study methodology. Exergaming and hippotherapy were excluded as systematic reviews on these topics recently have been performed for children with ADHD (Helmer, Wechsler, & Gilboa, 2021; Peñuelas-Calvo et al., 2022; White, Zippel, & Kumar, 2020).
3. *Outcome*: Treatment effect was determined with a standardized, reliable and valid developmental motor scale (e.g. 2nd edition of the Movement Assessment Battery for Children (MABC-2)).
4. *Design and language*: Only Controlled Clinical Trial (CCT) or Randomized Controlled Trial (RCT) designs written in English, Dutch, German or French were included. The references of systematic reviews and meta-analyses reporting on treatment in children with ADHD were screened.
5. *Publication date*: Studies published after the year 2000 were of interest, because of the publication of the DSM, text-revision of the 4th edition, giving more specified diagnostic criteria for ADHD and childhood specific classification factors.

2.4. Methodological quality

Two reviewers (LK, EV) independently applied The Physiotherapeutic Evidence Database (PEDro)-scale to assess methodological quality of the articles (total score: /10 (Cashin & McAuley, 2020)). Points were only awarded when a literal reading of the article clearly satisfied the criterion. Discrepancies were deliberated until consensus was reached.

Methodological quality was considered good if the total score was equal to or higher than 6, fair when equal to 4 or 5 and poor when equal to 3 or less (Cashin & McAuley, 2020).

2.5. Data extraction and synthesis of results

Information regarding the *population* (number of participants, dropouts, sample size, age, sex-distribution, diagnostic criteria, medication intake, current behavioral management and comorbidity), *interventions* (description, control intervention, rationale, total duration of the intervention, frequency and duration per session in accordance with the template for intervention description and replication (TIDieR) checklist (Hoffmann et al., 2014)), *outcomes and timepoints* (standardized developmental motor scales; pre-post assessments) and *results* (pre-test scores, post-test scores, statistical tests for comparison, significance level) was extracted from each individual study by one reviewer and its accuracy was checked by another (LK or EV). The motor-based interventions were grouped based on their composition and underlying rationale.

2.6. Data-analysis

Random-effects meta-analyses were calculated using a self-developed Excel file to estimate the pooled standardized mean differences (SMD) in motor skill performance after motor-based interventions. Meta-analysis was conducted if at least five studies were available (Jackson & Turner, 2017). To be included in the meta-analysis studies should report raw total scores of the standardized motor scale and the groups should be similar at baseline regarding the outcome measures of interest.

First, we calculated SMDs at individual study level with Hedges' g (Appendix S2-Formula 1) (Lee, 2016). SMDs can be interpreted as small (0.2), medium (0.5) or large (0.8) (Lee, 2016). In order to maximize the data input for the pooled outcome measures, we implemented the post-test values in preference to the changes from baseline. Because not all studies reported pretest comparison between groups, we first compared the pretest values using Hedges' g. Studies were only included in the meta-analysis if there were no baseline differences between the groups based on Hedges' g confidence interval.

Next, heterogeneity was determined by calculating the Q-statistic (Appendix S2-Formula 2) and I^2 (Appendix S2-Formula 3), representing the percentage of variation across studies that is due to heterogeneity rather than chance (Higgins & Thompson, 2002). Higher I^2 values indicate more heterogeneity among individual studies (Higgins & Thompson, 2002). If heterogeneity was too large (>50%), we planned subclassification based on the type of intervention. The pooled SMD, w (Appendix S2-Formula 4), was estimated using the within study variance (SE) and between study variance (tau-squared, t^2), and was expressed as a mean value and its 95% CI.

2.7. Quality of evidence

For studies comparing motor-based therapy to a non-motor control, the quality of evidence was appraised according to the Grading of Recommendation, Assessment, Development and Evaluation (GRADE)-method (Balshem et al., 2011). Information on the study limitations, imprecision, inconsistency, indirectness and publication bias was used in the quality assessment (Balshem et al., 2011). Because of the large heterogeneity, the quality of evidence assessment was not performed for studies comparing different types of motor-based interventions. Two investigators (LK, EV) independently conducted the quality of evidence assessment. Any discrepancy was discussed until consensus was met.

3. <u>Results</u>

3.1. Study selection

The literature search revealed 380 records. Thirteen studies (including 399 participants with ADHD) were eligible for methodological quality assessment and data-extraction as shown in the PRISMA-flowchart (Figure S3). One study was excluded because the applied intervention was insufficiently described to allow for data-extraction and overall interpretation of the results (O'Connor et al., 2014).

3.2. Methodological quality of individual studies

Only one of the included studies had good methodological quality (PEDro-score ≥ 6) (Pan et al., 2017). Six RCTs (Da Silva et al., 2020; Meßler, Holmberg, & Sperlich, 2018; Orangi, Yaali, Ghorbanzadeh, Loprinzi, & Ebdalifar, 2021; Pan et al., 2017; Pan et al., 2016; Yazd, Ayatizadeh, Dehghan, Machado, & Wegner, 2015) and three CCTs (Chang, Hung, Huang, Hatfield, & Hung, 2014; Pan et al., 2019; Verret, Guay, Berthiaume, Gardiner, & Béliveau, 2012) had moderate methodological quality, while one RCT (Ziereis & Jansen, 2015) and three CCTs (Jung & Suh, 2017; Kosari, Hemayattalab, Ameri, & Keihani, 2012; Torabi, Farahani, Safakish, Ramezankhani, & Dehghan, 2018) had low methodological quality. Table S4 presents detailed item scores for the individual studies.

3.3. Participant characteristics

Table 1 describes the participant characteristics. The sample sizes per group ranged from 8 (Jung & Suh, 2017) to 25 (Torabi et al., 2018), with an overall mean age of 9.98 (SD of the means: 1.4). Although all participants were diagnosed with ADHD, the method of diagnosis was not clearly described in two of the included studies (Jung & Suh, 2017; Kosari et al., 2012). Three studies (Pan et al., 2017; Pan et al., 2016; Pan et al., 2019) described the presence of comorbid neurodevelopmental disorders (i.e. Autism Spectrum Disorder (ASD), Tourette syndrome and Oppositional Defiant Disorder), while eight (Chang et al., 2014; Da Silva et al., 2020; Orangi et al., 2021; Pan et al., 2019; Torabi et al., 2018;

Verret et al., 2012; Yazd et al., 2015; Ziereis & Jansen, 2015) defined clear exclusion criteria in terms of (neurodevelopmental) comorbidities. Three studies (Jung & Suh, 2017; Kosari et al., 2012; Meßler et al., 2018) did not apply any exclusion criteria on comorbid disorders, nor described their presence. Eight studies reported the number of participants that were taking medication at baseline (Chang et al., 2014; Meßler et al., 2018; Pan et al., 2017; Pan et al., 2016; Pan et al., 2019; Verret et al., 2012; Yazd et al., 2015; Ziereis & Jansen, 2015), one of which considered medication intake as a randomization criterion (Pan et al., 2016). One additional study reported that medication intake was not altered throughout the intervention period, without presenting data on the number of participants that were taking medication (Da Silva et al., 2020). The medication of interest was specified in four studies only (Chang et al., 2014; Verret et al., 2012; Yazd et al., 2015; Ziereis & Jansen, 2015). Furthermore, except for Yazd et al. (2015), none of the included studies controlled for medication intake throughout the intervention period, nor reported medication dosage. Two studies reported an equal amount of sports activities at baseline (Meßler et al., 2018; Ziereis & Jansen, 2015), but none clearly described other services and physical activities delivered to the participants throughout the intervention period.

INSERT TABLE 1

3.4. Outcome measures

Treatment efficacy (Table 2) was objectified using the MABC-2 (Meßler et al., 2018; Ziereis & Jansen, 2015), the 2nd version of the Bruininks-Oseretsky Test of Motor Proficiency (BOT-2) (Kosari et al., 2012; Orangi et al., 2021; Pan et al., 2017; Pan et al., 2016; Torabi et al., 2018; Yazd et al., 2015), the 2nd version of the Test for Gross Motor Development (TGMD-2) (Pan et al., 2019; Verret et al., 2012), the Körperkoordinationstest für Kinder (KTK) (Da Silva et al., 2020; Jung & Suh, 2017) or the Basic Motor Ability Test (BMAT) (Chang et al., 2014). Raw group scores are presented in Table S6.

Ten studies defined motor skills as a primary outcome measure (Da Silva et al., 2020; Jung & Suh, 2017; Kosari et al., 2012; Meßler et al., 2018; Orangi et al., 2021; Pan et al., 2017; Pan et al., 2016; Pan et al., 2019; Torabi et al., 2018; Yazd et al., 2015) and three as a secondary outcome measure (Chang et al., 2014; Verret et al., 2012; Ziereis & Jansen, 2015) in addition to core ADHD symptoms (Chang et al., 2014; Verret et al., 2012), executive/cognitive functioning (Verret et al., 2012; Ziereis & Jansen, 2015) and physical fitness (Verret et al., 2012).

INSERT TABLE 2

3.5. Motor-based interventions for children with ADHD

3.5.1 Types of interventions

Table S5 contains information on the content of the interventions, their rationale, modes of delivery and how they were tailored to the needs and/or preferences of each child. Three intervention types were identified: 1) physical activity programs (PAP), as an umbrella term for interventions targeting physical activity and physical fitness by combining different kinds of motor activities; 2) motor skill training programs focusing on either fundamental motor skills (FMS training program - e.g. ball skills), or on sports-specific skills (e.g. table tennis); 3) perceptual-motor training, tapping into specific psychomotor functions such as body awareness and balance.

The *PAPs* are based on the rationale that physical activity and -fitness have a positive impact on executive functioning (Chang et al., 2014; Meßler et al., 2018; Verret et al., 2012), motor skills (Kosari et al., 2012; Pan et al., 2017), severity of ADHD symptoms (Torabi et al., 2018) and participation in physical activities (Orangi et al., 2021), whereas the *motor skill training programs* mainly focus on the link between motor skills mastery and executive functioning (Da Silva et al., 2020; Pan et al., 2016; Pan et al., 2019; Ziereis & Jansen, 2015). *The perceptual-motor interventions* identified in this review were based on the rationale that presenting a wide range of experiences involving motor and sensory tasks positively impacts motor skills, psychological functioning, social functioning and material experience (Jung & Suh, 2017; Yazd et al., 2015).

In general, the modes of delivery and tailoring of the interventions were seldomly described in the included studies. In five studies, the motor-based interventions were organized as group therapy, with the student-teacher ratio ranging from 6:3 (Pan et al., 2017) to 10:1 (Chang et al., 2014). In two studies, the participants were coached individually (Pan et al., 2016; Pan et al., 2019). Eight studies did not provide any information on the practical organization of the intervention. Tailoring of the intervention, in terms of intensity, was based on the heart rate in two studies (Meßler et al., 2018; Torabi et al., 2018), both including a PAP intervention. Only three studies (Pan et al., 2017; Pan et al., 2016; Pan et al., 2019) adapted the complexity of the exercises to the motor skill level, individual needs and/or preferences of the children.

3.5.2 Training volume

The training volumes per individual study are shown in Table 2. On average, the interventions lasted 9 weeks, ranging between 3 (Meßler et al., 2018) to 12 weeks (Jung & Suh, 2017; Pan et al., 2017; Pan et al., 2016; Pan et al., 2019; Ziereis & Jansen, 2015). The frequency varied from 1 (Pan et al., 2017; Ziereis & Jansen, 2015) to 3 sessions per week (Meßler et al., 2018; Orangi et al., 2021; Torabi et al., 2018; Verret et al., 2012; Yazd et al., 2015). One study (Kosari et al., 2012) did not describe the frequency and period of intervention, two other studies (Torabi et al., 2018; Yazd et al., 2015) did not report the duration of each individual therapy session.

3.5.3 Effectiveness of the interventions

Seven studies (Chang et al., 2014; Kosari et al., 2012; Meßler et al., 2018; Orangi et al., 2021; Pan et al., 2017; Torabi et al., 2018; Verret et al., 2012) investigated the effectiveness of different PAPs, four of motor skill training programs (Da Silva et al., 2020; Pan et al., 2016; Pan et al., 2019; Ziereis & Jansen, 2015) and two of perceptual-motor interventions (Jung & Suh, 2017; Yazd et al., 2015).

Motor-based interventions versus any non-motor control intervention

Five studies, constituting data from 146 children with ADHD, were suitable for meta-analysis (Jung & Suh, 2017; Pan et al., 2017; Pan et al., 2016; Torabi et al., 2018; Yazd et al., 2015). When comparing any type of motor-based intervention (PAPs (Pan et al., 2017; Torabi et al., 2018), motor skill training programs (Pan et al., 2016) or perceptual-motor interventions (Jung & Suh, 2017; Yazd et al., 2015)) to any non-motor control intervention (no intervention or medication), resulted in a large positive effect (pooled mean:1.46; 95% CI=[1.00;1.93]; I^2=47.07%) on motor skill performance in children with ADHD, regardless of the applied outcome measure (Figure 1).

INSERT FIGURE 1

In addition to the meta-analysis, five studies also reported effects of motor-based interventions compared to no intervention: two PAPs (Chang et al., 2014; Verret et al., 2012) and three motor skill training programs (Da Silva et al., 2020; Pan et al., 2019; Ziereis & Jansen, 2015). Three of these studies did not report raw data (Pan et al., 2019; Verret et al., 2012; Ziereis & Jansen, 2015) and two reported item scores only (Chang et al., 2014; Da Silva et al., 2020), as such these studies were not included in the meta-analysis. Gross motor skills, measured with the TGMD-2 (p=0.007) (Verret et al., 2012) and BMAT (target throwing (p=0.01) and bead moving (p=0.04) (Chang et al., 2014)), improved significantly more in the PAP groups (i.e. moderate-to-vigorous intensity training (Verret et al., 2012) and an aquatic exercise program (Chang et al., 2014) respectively) compared to no intervention.

A swimming learning intervention led to significantly better results on one KTK task, i.e. the side jumps (p=0.041) (Da Silva et al., 2020), whereas a table tennis program focusing on skill acquisition and executive functioning resulted in improved TGMD-2 results (Pan et al., 2019). Motor skill training programs with either high or low demands on manual dexterity, ball skills and balance, did not result in better performances on the MABC-2 than when no intervention was given (Ziereis & Jansen, 2015).

Comparing different motor-based interventions to each other

Not enough data were available to perform a meta-analysis per motor-based intervention. When comparing different PAPs in children with ADHD, diverging results were found. A high-intensity interval PAP significantly improved manual dexterity (p=0.045), but not balance (p=0.730) and ball skills (p=0.154) (MABC-2) compared to another PAP during which ball and team games, court sports and climbing were trained, only improving ball skills (p=0.025) (Meßler et al., 2018). A structured PAP, using sports, play and active recreation was more effective in improving the BOT-2 subtests (p-values not reported), compared to routine activities (PE classes) (Kosari et al., 2012). One study compared PAPs using aerobic exercises during which different learning pedagogies were used (Orangi et al., 2021). Based on their results a non-linear learning pedagogy, during which no feedback on task performance is given, seems more effective to improve fine and gross motor skills (BOT-2, no p-values reported).

One study (Yazd et al., 2015) investigated the effectiveness of a perceptual-motor intervention to improve motor skill performance in children with ADHD, showing that perceptual-motor training alone was equally effective in improving fine and gross motor skills, measured with the BOT-2 (p=0.591), compared to an identical dose of perceptual-motor training combined with drug therapy.

Only one study compared different types of motor-based intervention (i.e. PAP vs. FMS training). Their results showed similar improvements on the MABC-2 after motor skill programs with low or high demands on manual dexterity, ball skills and balance.

3.6. Quality of evidence

There is a low quality of evidence in favor of motor-based interventions compared to a non-motor control (Table 3), based on consistent results in low to moderate quality research. The estimated effect is likely to change based on further research.

INSERT TABLE 3

4. <u>Discussion</u>

This review aimed to determine the effectiveness of motor-based interventions on motor skill performance in children with ADHD. Strong evidence in literature is currently lacking, shown by only 13 relevant studies, all using different training modalities and outcome measures. This lack of evidence compromises the development of clinical guidelines on motor-based interventions in these children. Despite the differences in the rationale and thus the type of intervention, any type of motor-based intervention is better than a non-motor control to improve motor skills in children with ADHD, as shown by our meta-analysis. This result is in accordance with a recent meta-analysis, showing that increased physical exercise improves motor skills (Sun et al., 2022). As their meta-analysis only included four studies, their results should be interpreted with caution.

Moreover, additional high quality RCTs that replicate this finding are needed to increase GRADE confidence. Evidence for the superiority of a specific type of motor-based intervention is currently lacking, due to a very limited number of studies. Determining optimal treatment parameters is currently impossible, as most studies only provide a limited description of relevant parameters such as delivery modes and tailoring of the intervention. Future studies should clearly describe the rationale of the intervention and training methodologies including training parameters following the TIDieR guidelines (Hoffmann et al., 2014).

Stimulating children with ADHD, who have a higher risk for developing obesity (Cortese & Tessari, 2017; Quesada, Ahmed, Fennie, Gollub, & Ibrahimou, 2018) to be more physically active, as obtained with the PAPs, will impact their physical fitness and weight status (Quesada et al., 2018; Stodden et al., 2008). Clinicians, PE instructors and policy makers can use these PAPs to stimulate an active lifestyle, eventually empowering these children to take control over their own situation. A previous review reported that systematic physical activity (≥30 min per day, ≥40% intensity, ≥three days per week, ≥five weeks) is needed to further improve executive functioning and motor control (Suarez-Manzano et al., 2018). Nevertheless, crucial information for making similar recommendations on the use of PAPs to improve motor skill performance is missing. Group training is beneficial not only for increasing physical activity, but also for the social aspect (Pan et al., 2016). However, information on how to implement this in the PAP, is still lacking: only two studies (Chang et al., 2014; Pan et al., 2016) reported the student-teacher ratio, and one study (Pan et al., 2017) tailored the lessons to the motor skill level and personal preference of the children. Thus, methodologies should be stated clearer in future research if PAPs, shown to be effective, are to be successfully reproduced. Furthermore, there is a paucity of studies that investigate the effect of training intensity. Only one study compared high-intensity training to lower intensity training (Meßler et al., 2018), making it impossible to draw any conclusions on the optimal training intensity. Because the ultimate aim of motor-based interventions is to enhance participation in physical activities and empowering children to take control over their own situation, the program needs to contain motor skill activities, strength exercises, combined with an active lifestyle, i.e. at least 60 minutes of daily moderate-to-vigorous physical activity (Faigenbaum, MacDonald, Stracciolini, & Rebullido, 2020). However, this approach in children with ADHD is currently lacking in literature and deserves attention due to its importance for daily participation.

Providing more movement opportunities is not the same as specifically training motor skills based on requests for help as defined by the child, parents or other caregivers. Interventions targeting motor skill improvements should be based on conceptual frameworks, like motor learning paradigms (e.g. Neuromotor Task Training (Smits-Engelsman et al., 2018)). Such interventions use a task-oriented approach and are therefore meaningful for the child, goal-directed and focused on enabling the transfer of learning to the everyday context (Smits-Engelsman et al., 2018). Although a large body of evidence underpins the effectiveness of such motor learning paradigms, they have not been assessed in children with ADHD.

As a fundamental relationship between motor performance, social participation and underlying perceptual processes has previously been suggested (Chu & Reynolds, 2007), perceptual-motor interventions, including a wide range of sensorimotor tasks, might be a valuable type of motor-based intervention for children with ADHD. However, based on current literature, the effectiveness remains unclear.

Although the underlying mechanism explaining the effectiveness of motor-based intervention is not yet clearly defined, the importance of physical activity to promote neural development and neuroplasticity has been established (Halperin, Berwid, & O'Neill, 2014; Mandolesi et al., 2018). Various neuro-imaging studies indicate widespread structural (Halperin et al., 2014; Pereira-Sanchez & Castellanos, 2021; Saad, Griffiths, & Korgaonkar, 2020) and functional anomalies (Choi, Han, Kang, Jung, & Renshaw, 2015; De La Fuente, Xia, Branch, & Li, 2013; Rubia, 2018; Saad et al., 2020; Suskauer et al., 2008) in children with ADHD. While frontostriatal structures and frontal corticostriatal circuit abnormalities are most often described, an increasing number of studies additionally emphasize the involvement of other cortical regions (e.g. supplementary motor area and superior temporal lobe) and the cerebellum, thereby providing a plausible neurological explanation for the motor skill difficulties in children with ADHD. Future studies are needed to unravel the impact of motor-based interventions on the interplay between behavioral and neurological outcome measures.

4.1. Strengths and limitations of this study

Although the meta-analysis provides promising results, they should be interpreted in the light of this review's limitations and these of the included studies. More than one third of children the with ADHD have one or more comorbid disorder, such as DCD (Watemberg, Waiserberg, Zuk, & Lerman-Sagie, 2007) and/or ASD (Gnanavel, Sharma, Kaushal, & Hussain, 2019), which can have a significant impact on their motor skills. We aimed to study the effectiveness of motor-based interventions in children with ADHD, as such, studies on children with a comorbid diagnosis of DCD were excluded because of their known motor deficits. However, since three studies (Jung & Suh, 2017; Kosari et al., 2012; Meßler et al., 2018) did not consider comorbidities, the intended homogeneity cannot be guaranteed. Neglecting comorbidities can cause bias in the results and outcomes of the different studies. Therefore, future studies should provide a clear description of the study population, including comorbidities, to allow for subgrouping and eventually indicate disparity in the effect among different population groups. Although double blinding is hardly possible in motor-based interventions (Sherrington, Moseley, Herbert, Elkins, & Maher, 2010), other methodological aspects, such as concealed allocation, group similarity and blinding of assessors, can be controlled, but often were not. Also, not all outcome measures were specifically validated for the investigated population.

Because of the interrelatedness between physical activity, physical fitness and motor skills (Stodden et al., 2008), tackling one aspect seems to impact the others as well. However, when specifically training motor skills, motor skill performance is expected to be the primary outcome, whereas stimulating physical activity would improve physical activity and -fitness. Unfortunately, for several studies a mismatch in the type of training and the primary outcome measure was identified; the majority of the studies intended to improve motor skills by performing a PAP. As this type of intervention primarily addresses physical fitness and physical activity, the full potential of these interventions might be underestimated. Future studies should include measures of motor skill performance, physical fitness, and -activity to corroborate collective improvements among these factors.

The conflicting results underline the impact of *task-specificity* during training on the outcome. For instance, learning swimming techniques did not result in better KTK performance (Da Silva et al., 2020). Thus, future research should include both (non-) related outcome measures to explore intended effects but also carry-over effects to other skill domains.

The heterogeneity of the meta-analysis was borderline insignificant, based on the I^2 value of 47%. Because of the low number of included studies, looking at potential moderators of this heterogeneity was not possible. However, various study-level factors with the potential to contribute to the heterogeneity can be put forward, including the presence or absence of comorbid NDDs, the different outcome measures used, as well as differences in the applied interventions (e.g. intensity, duration, content).

4.2. Clinical implications and recommendations for future research

This research domain is relatively new and contains large knowledge gaps. Currently, any type of motor-based training is better than doing nothing in children with ADHD. It is essential to determine which type of training and which treatment methodologies are superior regarding impact on, among others, motor skill performance, functionality, participation in sports, quality of life, social interaction, cost-effectiveness and medication use (i.e. stimulants). Especially the latter needs further exploration. So far, only one study compared the effect of motor-based interventions to ADHD medication on motor skill performance, and though promising, new research is needed to confirm this finding. Furthermore, patient characteristics, such as age, comorbid (neurodevelopmental) disorders, ADHD subtype and baseline motor skill performance level, should be included in the analysis to determine the moderating effects of these individual characteristics and to improve homogeneity among different studies. Future research should include population groups with different combinations of comorbid disorders to disentangle the effectiveness of motor-based interventions in different (combined) pathologies (e.g. ADHD alone, combination of ADHD and ASD, ASD alone). Also, in terms of the motor-based interventions provided (e.g. type of intervention, intensity) and outcome measures used (e.g. construct of the outcome measure), more homogeneity is highly needed to identify the real potential of these interventions. Finally, future studies should focus on therapy tailored to the child's needs, preferences and skill level. Although the optimal therapy dose cannot be recommended, the importance of therapy adherence and at least a minimal amount of exercise is needed to obtain improvements in motor skill performance. Implementing the children's individual goals and working towards them, could assist in keeping the children motivated, enhancing an active lifestyle and preventing the onset of secondary problems such as overweight.

5. Conclusion

Evidence on the long-term consequences of motor skill deficits for children with ADHD, including overweight, sedentary lifestyle and social isolation, underpins the need for appropriate motor-based interventions and targeted outcome measures at different levels of biopsychosocial functioning. Based on current literature, motor-based interventions seem effective to improve motor skills in children with ADHD. However, no conclusion can be drawn on the most effective type of motor-based intervention and optimal treatment parameters. High quality RCTs comparing different types of motor-based interventions and different treatment methodologies in children with ADHD are required to determine the best choice of a specific type of motor-based therapy and to establish optimal treatment parameters.

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Tables

Table 1: Participant characteristics

Main author(s), year	Intervention groups	Participants: *Mean age (SD) (years)*	*N (dropouts)*	♂/♀	Diagnosis based on	Comorbidities (N)	Medication intake during intervention period (N)	Other services provided during intervention period (N)
Chang, 2014	Aquatic exercise program	8.8 (8.3)	15 (1)	10/4	DSM-IV-TR	Exclusion criterion: no neurological disorder	Methylphenidate (7)[#]	Behavioral treatment (3)[#]
	No intervention (wait-list)	8.2 (7.7)	15 (2)	13/0			Methylphenidate (6)[#]	Behavioral treatment (0)[#]
Da Silva, 2019	Swimming-learning training	12.0 (1.0)	18 (8)	6/4	DSM-IV-TR	Exclusion criterion: no comorbidities	Medication not altered during study[§]	/
	No intervention	12.0 (2.0)	15 (5)	8/2				
Jung & Suh, 2017	Material-experience perceptual-motor program	8-9*	8 (0)	8/0	/	/	/	/
	No intervention	8-9*	8 (0)	8/0				
Kosari, 2013	Physical education program based on SPARK	8.9 (0.8)	10 (?)	10/0	/	/	/	/
	Routine activities	8.8 (0.7)	10 (?)	10/0				
Meßler, 2018	High-intensity interval training	11.0 (1.0)	28 (?)	28/0	ICD-10	/	Unspecified medication (5)[#]	Routine sports therapy: 1-2x/week (28)[#] Identical amounts of psychotherapy, psychoeducation, counseling, behavioral management, ergotherapy, and music therapy for both groups[§]
	Ball and team games, court sports and climbing						Unspecified medication (4)[#]	
Orangi, 2021	Aerobic training (without feedback – non-linear learning pedagogy)	11.3 (1.0)	18 (?)	18/0	DuPaul's rating scale	Exclusion criteria: no other physical or psychological problems	/	/
	Aerobic training and other physical activities (with feedback – linear learning pedagogy)	11.3 (0.9)	18 (?)	18/0				
Pan, 2016	Table tennis program	8.9 (1.5)	16 (0)	16/0	DSM-IV-TR; CBCL	Asperger syndrome (1), Tourette syndrome (2), ODD (2)	Unspecified medication (9) + advised to maintain current pharmacological treatment	/
	No intervention	8.9 (1.6)	16 (0)	16/0			Unspecified medication (9) + advised to maintain current pharmacological treatment	
Pan, 2017	Physical activity training program	9.6 (2.5)	12 (0)	12/0	DSM-IV-TR; NVIQ	Asperger syndrome (1), Tourette syndrome (2), ODD (2)	Unspecified medication (15)[#]	Advised to maintain current behavioral management[§]
	No intervention	9.4 (2.7)	12 (0)	12/0				
Pan, 2019	Table tennis program	9.1 (1.4)	15 (0)	15/0	DSM-IV-TR; CBCL	ODD (2); Exclusion: ASD, ID, and CP	Unspecified medication (9) + advised to maintain current pharmacological treatment	/
	No intervention	8.9 (1.7)	15 (0)	15/0			Unspecified medication (9) + advised to maintain current pharmacological treatment	
Torabi, 2018	High-intensity intermittent training girls	12.6 (0.1)	15 (?)	0/15	DSM-IV; CPRS	Exclusion criteria: no cardiovascular, metabolic or respiratory diseases, neurological, intellectual or musculoskeletal disorders	/	/
	High-intensity intermittent training boys	12.5 (0.2)	10 (?)	10/0				
	No intervention girls	12.4 (0.5)	15 (?)	0/15				
	No intervention boys	12.6 (0.4)	10 (?)	10/0				
Verret, 2012	Moderate to vigorous intensity training program	9.1 (1.1)	10 (0)	9/1	DSM-IV-TR	Exclusion criterion: no other NDD	Methylphenidate (3) + advised to maintain current pharmacological treatment	Advised to maintain current behavioral management[§]
	No intervention		11 (3)	10/1			Methylphenidate (11) + advised to maintain current pharmacological treatment	
Yazd, 2015	Perceptual-motor training	8.7 (2.0)	12 (?)	10/2	DSM IV-TR; CPRS	Exclusion criteria: psychiatric or overt neurological symptoms and drug therapy other than methylphenidate and risperidone	Methylphenidate & risperidone (0)	/
	Drug therapy (methylphenidate & risperidone)	8.0 (1.9)	12 (?)	10/2			Methylphenidate & risperidone (12)	
	Perceptual-motor training + drug therapy	7.7 (1.3)	12 (?)	10/2			Methylphenidate & risperidone (12)	
Ziereis & Jansen 2014	Motor skill training program with high demands on manual dexterity, ball skills and balance	9.2 (1.3)	13 (0)	32/11	ICD-10	Exclusion criterion: no neurological disorders, no ASD, no mental retardation	Methylphenidate (0)	Equal amount of activities a sports club and leisure sports before testing[#]
	Motor skill training program using sports with low demands on manual dexterity, ball skills and balance	9.6 (1.6)	14 (0)					
	No intervention (wait-list)	9.5 (1.4)	16 (0)					

Explanations: * No age reported, age range as defined in inclusion criteria presented; (?) Dropouts not reported or not specified out of which group children dropped out; / Not clearly reported; [#] Not reported if medication/other services were discontinued during intervention period; [§] No data reported

Abbreviations: ***SD***=Standard deviation; ***DSM-IV-TR***=Diagnostic and Statistical Manual of Mental Disorders, 4th edition, text-revision; ***SPARK***=Sports, play and active recreation; ***ICD-10***=International Classification of Diseases and related health problems, 10th version; ***CBCL***=Child Behavior Checklist; ***ODD***=Oppositional Defiant Disorder; ***NVIQ***=Non-verbal Intelligence Quotient; ***ASD***=Autism Spectrum Disorder; ***ID***=Intellectual Disability; ***CP***=Cerebral Palsy; ***NDD***=Neurodevelopmental Disorder; ***CRPS***=Conners Parent Rating Scale

Table 2: Treatment efficacy

Main author(s), year	Intervention groups	Training volume	Standardized outcome measure	Within group comparison – Time effect				Between group comparison	PEDro score
				IG1	IG2	CG	General		
Chang, 2014	IG: PAP-Aquatic exercise program CG: No intervention (wait-list)	*Frequency: 2/week* *Period: 8 weeks* *Duration: 90'/session*	**BMAT** *Target throwing* *Bead moving*	 * *				 IG>CG* IG>CG*	4
Da silva, 2019	IG: Sport-specific - Swimming-learning training CG: No intervention	*Frequency*: 2/week *Period*: 8 weeks *Duration*: 45'/session	**KTK** *Balance beam walking, transfer* *Single leg jump* *Side jumps*	 ns * *		 ns ns ns		 ns ns IG>CG*	4
Jung & Suh, 2017	IG: PM-Material-experience perceptual-motor program CG: No intervention	*Frequency*: 2/week *Period*: 12 weeks *Duration*: 60' (IG)	**KTK** *Balance beam walking, single leg jump, side jumps, transfer*					 IG>CG!	2
Kosari, 2013	IG: PAP-Physical activity training program IG2: PAP-Routine activities	*Frequency*: ? *Period*: ? *Duration*: 45'/session	**BOT-2** *Running speed and agility, balance, bilateral coordination, strength*	 ***	 ns			 IG>IG2!	2
Meßler, 2018	IG: PAP-High-intensity interval training IG2: PAP-Ball and team games, court sports and climbing	*Frequency*: 3/week *Period*: 3 weeks *Duration*: 30' (IG), 60' (CG)/session	**MABC-2** Total score *Manual dexterity* *Balance* *Ball skills*	 * * ns ns	 ns ns ns *			 IG>IG2* IG>IG2* ns	4
Orangi, 2021	IC: PAP-Aerobic training (without feedback – non-linear learning pedagogy) IG2: PAP-Aerobic training and other physical activities (with feedback – linear learning pedagogy)	*Frequency*: 3/week (IG), 2/week (CG) *Period*: 8 weeks *Duration*: 60'/session (IG), 90'/session (CG)	**BOT-2** Total score					 IG>IG2***	5
Pan, 2016	IG: Sport-specific - Table tennis program CG: No intervention (wait-list)	*Frequency*: 2/week *Period*: 12 weeks *Duration*: 70'/session	**BOT-2** Total motor proficiency score *Fine motor control, body coordination* *Manual coordination, strength & agility*				 ns ns ns	 IG>CG** ns IG>CG**	5
Pan, 2017	IG: PAP-Physical activity training program CG: No intervention	*Frequency*: 1/week *Period*: 12 weeks *Duration*: 90'/session	**BOT-2** Total motor proficiency score *Fine motor control, manual coordination, body coordination, strength & agility* *Fine motor precision, fine motor integration, upper-limb coordination, balance, run/agility, strength* *Manual dexterity, bilateral coordination*					 IG>CG** IG>CG** ns IG>CG**	6
Pan, 2019	IG: Sport-specific - Table tennis program CG: No intervention (wait-list)	*Frequency*: 2/week *Period*: 12 weeks *Duration*: 70'/session	**TGMD-2** *Locomotor skills (raw score)* *Object-control (raw score)*	 * *		 ** ns		 IG>CG** IG>CG**	4
Torabi et al. 2018	IG: PAP-High-intensity intermittent training CG: No intervention	*Frequency*: 3/week *Period*: 6 weeks *Duration*: ?	**BOT-2** *Total score*	 *		 *		 IG>CG**	2
Verret, 2012	IG: PAP-Moderate to vigorous intensity training program CG: No intervention	*Frequency*: 3/week *Period*: 10 weeks *Duration*: 45'/session	**TGMD-2** Total raw score *Locomotor skills (raw score)* *Object-control (raw score)*					 IG>CG** IG>CG** ns	4
Yazd, 2015	IG1: PM-Perceptual-motor training IG2: PM-Combined perceptual-motor training and drug therapy CG: Drug therapy	*Frequency*: 3/week (IG1, 3), daily (IG2) *Period*: 6 weeks (IG 1, 3) *Duration*: ?	**BOT-2** Total score – gross motor Total score – fine motor	 * *	 * *	 * *		 IG1=IG2>CG*** IG1=IG2>CG**	5
Ziereis & Jansen, 2015	IG1: FMS-Motor skill training program with high demands on manual dexterity, ball skills and balance IG2: Sport-specific - Motor skill training program using sports with low demands on manual dexterity, ball skills and balance CG: No intervention (wait-list)	*Frequency*: 1/week *Period*: 12 weeks *Duration*: 60'/session	**MABC-2** Total score *Manual dexterity* *Ball skills, balance*				 *** ** ns	 ns ns ns	3

Explanations: * p<0.05; ** p<0.01; *** p<0.001; !Statistical analysis not reported; ***ns*** Not significant; ***general*** Difference in outcome between time points, not further specified

Abbreviations: ***PAP***=Physical activity program; ***PM***=Perceptual-motor therapy; ***BMAT***=Basic Motor Ability Test; ***IG***=Intervention Group; ***CG***=Control Group; ***KTK***=Körperkoördinationstest für Kinder; ***BOT-2***=Bruininks-Oseretsky test of Motor Proficiency, 2nd edition; ***MABC-2***=Movement Assessment battery for children, 2nd edition; ***TGMD-2***=Test for Gross Motor Development, 2nd edition

Table 3: Quality of evidence appraised using the Grading of Recommendations, Assessment, Development and Evaluation (GRADE)-method

<table>
<tr><th>Study Design: No. of studies, risk of bias (ref.)</th><th>Study limitations</th><th>Consistency</th><th>Directness</th><th>Precision</th><th>Publication bias</th><th>Quality of the evidence (GRADE)</th></tr>
<tr><td>RCT: 1 low (Pan et al., 2017), 5 moderate (Da Silva et al., 2020; Meßler et al., 2018; Orangi et al., 2021; Pan et al., 2016; Yazd et al., 2015), 1 high (Ziereis & Jansen, 2015)</td><td rowspan="2">Some limitations (↓)</td><td rowspan="2">Consistent (=)</td><td rowspan="2">Some indirectness (↓*)</td><td rowspan="2">Some imprecision (=*)</td><td rowspan="2">No publication bias assumed (=)</td><td rowspan="2">⊕⊕⊖⊖</td></tr>
<tr><td>CCT: 3 moderate (Chang, 2014; Verret, 2012; Pan, 2019), 3 high (Jung & Suh, 2017; Kosari et al., 2012; Torabi et al., 2018)</td></tr>
</table>

Explanation: ↓GRADE score downgraded by one point; = No impact on GRADE score; * Based on heterogeneity among the included studies (e.g. presence of absence of comorbid NDDs, medication use, other services provided), there was some uncertainty about downgrading the evidence because of indirectness. To compensate the borderline decision, the precision category was not downgraded, despite some imprecision due to the small sample sizes of the included studies, as recommended by the GRADE guidelines.; ⊕ Point on final GRADE score awarded; ⊖ Point on final GRADE score not awarded

Abbreviations: ***RCT***=Randomized controlled trial; ***CCT***=Controlled clinical trial

GRADE Working Group grades of evidence: ***High quality***=Further research is very unlikely to change our confidence in the estimate of effect; ***Moderate quality***=Further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate; ***Low quality***=Further research is very likely to have an important impact on our confidence in the estimate of affect and is likely to change the estimate; ***Very low quality***=We are very uncertain about the estimate

Figures

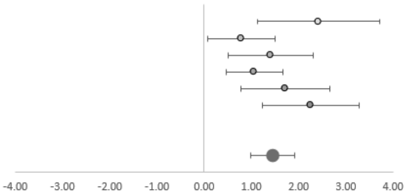
Figure 1: Forest plot for meta-analysis regarding the general effect of motor-based intervention on motor skill performance compared to no intervention or medication

Figure 1

Author (year)	Intervention (comparison)	Outcome Measure	Intervention group				Control group				Standardized mean difference (random effects model)		
			Age Group	*N*	*Mean*	*SD*	*Age Group*	*N*	*Mean*	*SD*	*Hedges' g*	*lower bound*	*upper bound*
Jung & Suh, 2017	PM - no intervention	KTK - total score		8	125.40	8.50		8	99.40	11.50	2.43	1.14	3.72
Pan, 2016	SS - no intervention	BOT-2 - total score	8.9 (1.5)	16	64.13	9.50	8.9 (1.6)	16	55.88	10.74	0.79	0.07	1.51
Pan, 2017	PAP - no intervention	BOT-2 - total score	9.6 (2.5)	12	57.25	10.86	9.4 (2.7)	12	44.50	5.84	1.41	0.52	2.31
Torabi, 2018	PAP - no intervention	BOT-2 - total score	12.6 (0.2)	25	41.92	9.42	12.5 (0.5)	25	33.42	5.84	1.07	0.47	1.66
Yazd, 2015	PM - medication	BOT-2 - gross motor total score	8.7 (2.0)	12	52.1	12.63	8.0 (1.9)	12	32.08	9.67	1.72	0.78	2.65
Yazd, 2015	PM - medication	BOT-2 - fine motor total score	8.7 (2.0)	12	42	2.33	8.0 (1.9)	12	28.33	7.96	2.25	1.23	3.27
Pooled mean	**PAP/PM/SS vs no intervention/mediation**	**standardizes motor scales - total scores**									**1.46**	**1.00**	**1.93**

Test of homogeneity, Q = 9.45, p=0.09; I^2=47.07%

Forest plot



Description: Post test scores are reported as described in the original articles
Abbreviations: ***SD***=Standard deviation; ***PM***=Psychomotor therapy; ***PAP***=Physical activity program; ***SS***=Sport-specific intervention; ***KTK***=Körperkoördinationstest für Kinder; ***BOT-2***=Bruininks-Oseretsky test of Motor Proficiency, 2nd edition