Evidence for increasing physical activity in children with physical disabilities: a systematic review

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ABBREVIATIONS

AACPDM	American Academy for Cerebral Palsy and Developmental Medicine
ICF	International Classification of Functioning, Disability and Health
RCT	Randomized controlled trial

AIM To summarize the best evidence of interventions for increasing physical activity in children with physical disabilities.

METHOD A systematic review was conducted using an electronic search executed in Academic Search Elite, Academic Search Premier, CINAHL, Embase, MEDLINE, PEDro, PsychINFO, and SPORTDiscus up to February 2016. The selection of articles was performed independently by two researchers according to predetermined eligibility criteria. Data extraction, methodological quality, and levels of evidence were independently assessed by two researchers using a data-collection form from the Cochrane Collaboration and according to the guidelines of the American Academy for Cerebral Palsy and Developmental Medicine.

RESULTS Seven studies were included. Five randomized controlled trials ranged from strong level I to weak level II studies, and two pre–post design studies were classified as level IV. There is level I evidence for no effect of physical training on objectively measured physical activity, conflicting level II evidence for interventions with a behavioural component on the increase of objectively measured physical activity directly after the intervention, and level II evidence for no effect during follow-up. Results are limited to children with cerebral palsy as no other diagnoses were included.

INTERPRETATION Increasing physical activity in children with physical disabilities is very complex and demands further development and research.

Increasing physical activity in children and adolescents is a subject of great interest in paediatric research, with alarmingly growing rates of obesity and inactivity. A physically active lifestyle is known to achieve fundamental health benefits for all individuals, including improved health-related quality of life, enhanced psychological well-being, improved physical functioning, and prevention of non-communicable disease in both adults and children.[1–5] While being active is important for typically developing children, children and adolescents with physical disabilities could benefit even more from regular physical activity in the prevention not only of comorbidity but also of functional decline and fatigue.[6–11] Despite these obvious benefits of physical activity, children with a physical disability are known to be at higher risk of an inactive lifestyle.[12–17]

Secondary to an inactive lifestyle, children with physical disabilities show reduced levels of cardiorespiratory fitness.[18,19] Therefore, several studies have evaluated the effects of training in children and adolescents with physical disabilities. Although these studies have shown positive results in measures of fitness, they have also shown that the increased levels of fitness do not sustain.[20,21] This is not entirely surprising because these training programmes were mostly aimed at increasing bodily functions such as fitness, and were not aimed at increasing participation in physical activity. Given the benefits of physical activity in maintaining gains in fitness and other health benefits, however, it seems that sufficient levels of physical activity should also be a primary goal and outcome for paediatric practice to obtain durable effects. Increasing participation in physical activity requires behavioural change,[22] because it is more than a skill as defined by the International Classification of Functioning, Disability and Health (ICF).[23] Factors associated with physical activity in children and adolescents with disabilities, such as self-efficacy and the presence of adaptive equipment, should ideally be incorporated into interventions aimed at increasing physical activity.[24–26] A recent paper has described the unique position of paediatric

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physical therapists in implementing interventions to achieve healthy, active lifestyles in children and adolescents.[27]

Although the evidence for the effectiveness of interventions aimed at improving physical activity is still limited in typically developing children,[28,29] the knowledge about children with physical disabilities seems even more limited and an overview of effective interventions is lacking. Therefore, the aim of this systematic review is to summarize the best evidence of interventions for increasing physical activity in children with physical disabilities. The results of this review will give more insight into effective ways of improving physical activity in paediatric rehabilitation practice.

METHOD

Search strategy

We conducted a systematic literature search up to and including February 2016 in the following electronic databases: MEDLINE, CINAHL, Academic Search Elite, Academic Search Premier, Embase, PEDro, PsycINFO, and SPORTDiscus. In consultation with a medical information specialist (JM), a comprehensive search strategy was developed around three major themes: 'children', 'disability', and 'physical activity'. Key terms within the search strategy were mapped to medical subject headings in MEDLINE and expanded to include narrower terms. Also, title and abstract search words were added to find the most recent, non-indexed literature. The MEDLINE search string was translated for the other databases. The complete search strategy for MEDLINE is presented in Appendix S1 (online supporting information). Finally, the reference lists of included studies were manually searched for additional studies.

Selection process and eligibility criteria

Titles, abstracts, and full texts were independently reviewed for eligibility by two reviewers (Manon Bloemen with Leontien van Wely or Manon Bloemen with Hanneke Borst or Leontien van Wely with Joyce Benner), with any discrepancies being discussed with a third reviewer (J. de Groot) until consensus was reached. Quantitative intervention studies were included if they had the following criteria: (1) the study participants were children (4–18y, or a group mean age <18y) with a physical disability; (2) the studies were randomized controlled trials (RCTs), clinical controlled trials, or single-group designs; (3) the interventions were part of paediatric physical therapy (such as physical training or exercising, task-oriented or functional training, interactive video gaming, promoting physical activity, coaching, motivational interviewing); (4) one of the reported outcome measures was either objectively or subjectively assessed physical activity; (5) it was a full-text peer reviewed article; (6) the studies were written in English or Dutch.

Studies were excluded if more than 50% of the participating children or adolescents did not have a physical disability and the results were not presented separately; the physical disability was of a progressive nature or if conditions were present in which exacerbations could occur; the study concerned single case reports or case series.

What this paper adds

- Physical training alone does not increase physical activity in young people with cerebral palsy.
- Behavioural interventions show conflicting evidence directly after the intervention.
- Behavioural interventions show no increase in physical activity at follow-up.

Data extraction

The data extraction was always performed by two independent reviewers (Manon Bloemen with Leontien van Wely or Manon Bloemen with Hanneke Borst), using an adapted data collection form from the Cochrane Library.[30] Any discrepancies between reviewers were resolved by discussion until consensus was reached. The data were incorporated in a summary table according to the guidelines of the American Academy for Cerebral Palsy and Developmental Medicine (AACPDM).[31]

Levels of evidence and methodological quality assessment of the papers

The levels of evidence and methodological quality according to the AACPDM were independently assessed by two researchers and any discrepancies were resolved by discussion until consensus was reached.[31] The levels of evidence of the AACPDM are based on those by Sacket[32] and range from level I (definite conclusions) to level V (no definite conclusions), as presented in Table I. The studies that were classified as levels I to III were assessed for methodological quality by seven criteria (see Table SI, online supporting information). The ratings for methodological quality range from strong (six or seven criteria positive) to moderate (four or five criteria positive) to weak (zero to three criteria positive).[31] Studies classified as levels of evidence IV or V are not rated for methodological quality because of threats of internal validity owing to weak study design.[31]

Strength of the evidence

Owing to the heterogeneity of the studies, a meta-analysis could not be performed. The outcomes, measures, and statistical results of the studies classified as levels of evidence

Table I: Levels of evidence for studies[31]

Level	Intervention (group) studies
I	Systematic review of randomized controlled trials (RCTs) Large RCT (with narrow confidence intervals [n>100])
II	Smaller RCTs with (with wider confidence intervals [n<100]) Systematic reviews of cohort studies 'Outcome research' (very large ecological studies)
III	Cohort studies (must have concurrent control group) Systematic reviews of case–control studies
IV	Case series Cohort study without concurrent control group (e.g. with historical control group) Case–control study
V	Expert opinion Case study or report Bench research Expert opinion based on theory or physiological research Common sense/anecdotes

I to III are presented and analysed as recommended by the AACPDM guidelines.[31] According to these guidelines, the results should be classified according to the ICF into body structures/body functions, activities and participation, and contextual factors.[31]

RESULTS

Search results

The electronic search resulted in 6071 studies. After screening titles and abstracts, 6001 studies were excluded. The full texts of the remaining 70 studies were analysed; of these, another 62 were excluded, mostly because they did not report effects on physical activity. One additional study[33] was excluded because it was a feasibility study and did not have the objective to measure the effect of the intervention. The full texts of the remaining seven studies[34–40] were reviewed, summarized in a data extraction form, and included in this systematic review. Five[35–38,40] of these seven studies were RCTs, and two[34,39] used a single-group design. One study[38] was an RCT during the first year of the trial and continued with a single-group design during the second year.

A manual search of the reference lists of the included studies identified no additional studies. The selection process is presented in Figure S1 (online supporting information).

Data extraction

The characteristics of the studies are reported in Table SII (online supporting information). All seven studies[34–40] included children with cerebral palsy (CP) at Gross Motor Function Classification System levels I to II,[40] I to III,[34–37] and I to V,[38,39] with ages ranging from 6 (Crompton et al.[35]) to 17 (Mitchell et al.[40]) years. Sample sizes ranged from 15 (Crompton et al.[35]) to 102 (Mitchell et al.[40]).

Levels of evidence and methodological quality

The levels of evidence of the studies varied from level I[40] to level II[35–38] to level IV[34,38,39] and is reported in Table SII. The first part of the study from van den Berg-Emons et al.[38] was classified as an RCT and the last part as a pre–post design. The methodological quality of the included studies classified as levels of evidence I to III is reported in Table SI. One level I RCT[40] and one level II RCT[37] were rated as having strong methodological quality. Two level II RCTs were rated as having moderate methodological quality[36,38] and one as having weak methodological quality.[35]

Content of the interventions of all included studies

There was large variation in the content of the interventions. Five studies focused on physical training[34,35,38–40] such as strengthening, endurance training, and balance training. The training volumes varied tremendously, from 20 hours a week for 3 weeks,[34] to 3 hours per week for 9 months.[38] All these studies[35,38–40] continued the usual therapy programme during the intervention, except for Christy et al.[34] The two other studies[36,37] focused on a more behavioural[36] or combined behavioural and physical training[37] approach. Maher et al.[36] used a highly interactive Internet-based programme, 'Get Set', based on cognitive theory, whereas Van Wely et al.[37] used a face-to-face lifestyle intervention, in which they combined group fitness with individual counselling and individual home-based physical therapy. The total length of the 'Get Set' programme was 8 weeks and usual physical therapy was continued,[36] whereas the lifestyle intervention of Van Wely et al.[37] lasted for 6 months.

Outcome measures of physical activity of the studies classified as levels of evidence I to III

The outcome measures of the five studies classified as levels of evidence I and II are presented in Table SIII (online supporting information). Within the focus of this review, the results on physical activity are described under 'activity and participation' according to the ICF.[23] All studies[35–38,40] reported measurements of physical activity before and directly after the intervention, with three studies[35–37] also measuring physical activity during follow-up, ranging from 6 weeks (Crompton et al.[35]) to 6 months (Van Wely et al.[37]). Four studies[35–37,40] measured objective physical activity by activity monitors; however, a variety of instruments were used and the wear time of the activity monitors varied from 4 days (Crompton et al.;[35] Mitchell et al.[40]) to 1 week (Maher et al.;[36] Van Wely et al.[37]). Moreover, a wide variety of outcome measures from the activity monitors were used. Crompton et al.[35] defined physical activity as uptime using a positional activity logger. Other activity monitors were used to report step counts,[36,37,40] time in light activity,[40] time in moderate to vigorous physical activity (moderate to vigorous physical activity),[36,40] weekly walking distance,[36] activity counts,[40] time spent at medium-to-high stride rate,[37] and time spent at high stride rate.[37] Van den Berg-Emons et al.[38] measured physical activity objectively using total energy expenditure.

Two studies[36,37] measured subjective physical activity. Maher et al.[36] used the MARCA, a self-reported physical activity questionnaire about four previous days, and Van Wely et al.[37] used the parent-reported, child-adapted AQuAA, with questions about the frequency and duration of physical activity during the previous 7 days. In addition, both studies used self-reported weekly[37] or daily[36] moderate to vigorous physical activity, self-reported daily level of physical activity,[36] or self-reported compliance with the physical activity norm.[37]

Effects of interventions on physical activity in the studies classified as levels of evidence I to III

Of the five RCTs[35–38,40] that measured objective physical activity, only one[36] found an effect for objectively measured physical activity directly after the intervention. It[36] reported an increase in weekly walking distance (effect size 0.96, p=0.05) in favour of the intervention group receiving the Internet-based programme 'Get Set' compared with usual care. No study reported significant differences during follow-up.[35–37]

Of the two RCTs[36,37] that measured subjective physical activity, only one[37] reported a significant difference at the end of the intervention for parent-reported time at moderate to vigorous physical activity in favour of the lifestyle intervention compared with usual paediatric physical therapy (between-group change ratio 2.2; 95% confidence interval 1.1–4.4; p=0.04). No significant differences were found at follow-up.[36,37]

Strength of the evidence

Looking at the content of the interventions of the studies classified as levels I to III, two groups of interventions can be identified: (1) interventions focusing on physical training alone[35,38,40] and (2) interventions using a behavioural component.[36,37] The strength of the evidence was separately analysed for these two groups of interventions and for objectively and subjectively measured physical activity. Results were limited to children with CP as no other diagnoses were included.

Physical training

There was one strong level I study,[40] one moderate level II study,[38] and one weak level II study[35] showing no effect, resulting in level I evidence for no effect of physical training on objectively measured physical activity in children with CP.

Behavioural component

There was one strong level II study[37] showing no effect and one moderate level II study[36] showing a significant effect, resulting in conflicting evidence for interventions with a behavioural component on the increase of objectively measured physical activity directly after the intervention. Both studies[36,37] found no effects at follow-up, resulting in level II evidence for no effect.

For subjectively measured physical activity, one strong level II study[37] showed a significant effect, and one moderate level II study[36] showed no effect, resulting in conflicting evidence for the increase of subjectively measured physical activity. As neither study[36,37] found effects at follow-up, there is level II evidence for no effects of interventions with a behavioural component on subjectively measured physical activity during follow-up.

DISCUSSION

The aim of this systematic review has been to summarize the best evidence of interventions for increasing physical activity in children with physical disabilities, in order to provide more insight into effective ways of improving physical activity in paediatric practice. In this search, only seven[34–40] studies were identified: four were RCTs,[35–37,40] two were single-group design studies,[34,39] and one[38] used an RCT design in the first part of the study and a single-group design in the last part. Five studies[35–38,40] were classified as level I to III studies according to the levels of evidence, and only results of these studies were included in weighing the strength of the evidence.

Interestingly, all studies included only children with CP. Compared with a systematic review from Bania et al.,[41] who analysed the effects of interventions on physical activity in people with CP and included both quantitative and qualitative studies, four additional studies[34,37,39,40] were included in our review. Bania et al.[41] concluded that structured exercise programmes and online behavioural programmes can be effective in increasing physical activity, but that the results are not maintained. Our review showed level I evidence for no effect of physical training on objectively measured physical activity in children with CP, conflicting evidence for the effect of interventions with a behavioural component on objectively and subjectively measured physical activity directly after the intervention, and level II evidence for no effect at follow-up. Unfortunately, the evidence in our review seems less positive than the conclusion of Bania et al.[41] several years ago. It was striking to learn that, even though increasing physical activity in children with physical disabilities is an important topic in paediatric practice, the knowledge base is still very small, and only evidence for children with CP was found.

We found a tremendous variation in the interventions that were applied. It was intriguing to notice that five studies used solely physical training[34,35,38–40] and thus focused on the level of bodily functions of the ICF. This focusing on bodily functions still seems to predominate goal setting and intervention in physical therapy practice. However, both literature[20,21] and studies included in this review[34,35,38–40] show this is insufficient in attaining goals at the level of participation, such as an increase in physical activity in daily life. This emphasizes the importance of shifting our focus from the level of bodily functions to the level of participation as defined by the ICF, and thus taking personal and external factors into account.[23]

Two of the included studies[36,37] integrated behavioural models in their interventions, with only Maher et al.[36] showing a significant difference in only one objective outcome measure of physical activity directly after the intervention. Unfortunately, this increase in physical activity was not sustained until follow-up. Change in participation involves a change in behaviour and, when aiming to change certain behaviours, models of behavioural change should be integrated into physical therapy practice. One of these models, specifically aimed at physical activity, is the model of physical activity for persons with a disability,[31] which combines the ICF with the model of attitude, social influence, and self-efficacy. In this model, the intention of being physically active is the central determinant for participation in physical activity, with aspects such as attitude, self-efficacy, health condition, and facilitators and barriers as personal components, and social influence and facilitators and barriers as external components. Using a combination of behavioural models and the ICF is also recommended by Johnston and Dixon.[42] These authors tested the prediction of behaviour using the ICF, behavioural models, and a combination of the ICF and behavioural models, and found that the combination performed best in predicting behaviour. Interestingly, these authors also hypothesized that non-volitional determinants such as impairments are more strongly related to measures of limitations and capacity, whereas volitional determinants are more strongly related to measures of performance, in this case participation in physical activity.[42] This is very much in line with recent studies[24,25] looking at factors associated with participation in physical activity. A focus on problem solving, self-efficacy, and the presence of positive personal and environmental factors seems crucial for participation in physical activity.[24,25] With this emphasis for combining behavioural models with the ICF in mind, what can we learn from the studies[36,37] using behavioural models in their interventions and why are the effects so marginal?

The ‘Get Set’ programme used by Maher et al.[36] was described as an 8-week ‘highly interactive Internet-based program based on social cognitive theory’. The retention rate during the trial was high, so why was there only an effect on one objective physical activity measure, and why was the increase in physical activity not maintained? An important aspect may be the duration of the programme, as it is known that behavioural changes take time to occur and it takes time and effort to truly maintain them.[43] Another essential aspect might be the fact that a ‘one size fits all’ programme may not be the answer, as each child and their parents experience specific facilitators and barriers for participation in physical activity that need to be individually addressed.[24,25] So, taking this individual approach into account, it is interesting to see that there were no significant findings for objectively measured physical activity in the RCT by Van Wely et al.,[37] despite their individually tailored approach. Their intervention consisted of motivational interviewing, together with fitness training and regular home-based physical therapy. Motivational interviewing is a client-centred interview style aimed at behavioural change, and all participants in this RCT received a minimum of three counselling sessions.

However, it is interesting to see that the attitudes towards, for example, sports of both the participating children and their parents were positive from baseline; the question arises whether motivational interviewing was the best treatment option for every participant or if another approach would have been more successful. The home-based physical therapy was aimed at increasing the capacity for daily activities; yet performance in physical activity is much more complicated than just capacity for daily activities, as shown by the wide variety of factors associated with physical activity.[24,25,44] So it seems that the content of the home-based physical therapy may not have focused enough on specific facilitators or barriers that might have been present and are part of the personal and environmental factors as described by both the ICF and the model of physical activity for persons with a disability. Furthermore, even though the whole intervention lasted for 6 months, this may still not have been long enough to indeed see behavioural change.[43]

Given the very limited evidence for effective interventions to improve physical activity in children with physical disabilities, it is very interesting to consider future directions. We believe that increasing physical activity levels in children with physical disabilities is very complex and needs an individual approach. The specific barriers and facilitators, which differ for every child because of differences in context and differences in personal factors, should be analysed thoroughly.[24,25] Clinical reasoning should then lead to an individual hypothesis about the causes of the reduced levels of physical activity. By doing so, the intervention that fits the hypothesis can be identified. Possible future interventions may be developed in co-design[45,46] with the children with physical disabilities and their parents. This is a relatively new approach that shows positive findings in other healthcare areas.[47–49] These new interventions should first be piloted in, for example, case studies or case series, after which they can be further developed and improved. If these stages are completed, larger effect studies may be undertaken to analyse the effect at a group level.

An important constraint is that outcome measures for assessing physical activity are limited in their clinimetric properties and feasibility. Both objective and subjective measures are used with results and are not interchangeable, owing to low agreement between the two methods.[50] Moreover, measuring physical activity in daily paediatric practice is still a challenge and not implemented within regular care yet.[51]

This systematic review used an extensive and sensitive search to identify all possible studies analysing the effects of interventions on physical activity in children with physical disabilities. One of its strengths was that the selection of studies, data extraction, and methodological quality assessment were performed by two independent reviewers. However, certain limitations should be taken into account when interpreting the results. For example only English and Dutch articles were included, so results from studies using another language may have been missed. Moreover, we chose to exclude qualitative studies, case reports, and case series because we wanted to report statistical differences at a group level. These qualitative studies, case reports, and case series may give insight, however, into new possibilities for increasing physical activity in children with physical disabilities. Another important aspect is that only children with CP were included in the studies, even though all non-progressive physical disabilities were allowed in this review, of course limiting the generalizability of our results.

CONCLUSION

In conclusion, there is level I evidence for no effect of physical training alone on objectively measured physical activity in children with CP, conflicting evidence for the effect of interventions with a behavioural component on objectively and subjectively measured physical activity direct after the intervention, and level II evidence for no effect at follow-up.

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SUPPORTING INFORMATION

The following additional material may be found online:

Appendix S1: Search terms.

Table SI: Methodological quality of the studies with level of evidence I–III

Table SII: Summary of studies: interventions and participants

Table SIII: Summaries of studies with level of evidence I–III: outcomes, measures, and results

Figure S1: Flow diagram detailing study selection process.

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