Title

A systematic review and meta-analysis of interventions to increase physical activity in children and adolescents with intellectual disabilities.

Abstract

Background: Increasing physical activity (PA) through intervention can promote physical and mental health benefits in children and adolescents. However, children and adolescents with intellectual disabilities (ID) have consistently been shown to engage in low levels of PA, which are insufficient for long-term health. Despite this, little is known about the effectiveness of interventions to increase PA in children and adolescents with ID. The aim of this study was therefore to systematically review how effective interventions are at increasing PA levels in children and adolescents with ID, and further examine what components have been used in these interventions.

Method: A systematic search of MEDLINE, EMBASE, ERIC, CINAHL, PsychINFO, CENTRAL, and ISRCTN trials registry was conducted (up to July 2016). Articles were included if they met the following eligibility criteria: children and adolescents (<18years) with ID, measurement of PA at baseline and post-intervention, intervention studies. Effect sizes were calculated as standardised mean difference (*d*) and meta-analysis calculated between intervention and no treatment control intervention.

Results: Five studies met the eligibility criteria and were included in the review. Study design, methodological quality, and intervention components were varied. Interventions did not support sufficient changes in PA to improve health. The meta-analysis demonstrated that intervention groups were not more effective at increasing PA levels post-intervention (*d*: 2.20; 95% CI -0.57 to 0.97) compared to control. However, due to a decrease in PA in the control intervention, a moderate significant effect was demonstrated at follow-up (*d*: 0.49; 95% CI 0.14 to 0.84).

Conclusions: There is a lack of studies which aim to increase PA levels in children and adolescents with ID, with current interventions ineffective. Future studies are required before accurate recommendations for appropriate intervention design and components can be made.

Key words

Systematic Review, Meta-analysis, Intellectual Disabilities, Children, Adolescents, Physical Activity, Intervention, Components.

Introduction

Physical activity (PA) is associated with many physical and mental health benefits in children and adolescents (Ahn & Fedewa, 2011; Biddle & Asare, 2011; Janssen & LeBlanc, 2010). However, recent studies report children and adolescents with ID to be inactive and not meeting the PA guideline of 60 minutes of moderate to vigorous intensity activity (MVPA) per day (Boddy et al. 2015; Einarsson et al. 2015). In comparison to their typically developing (TD) peers, children and adolescents with ID are less active and participate in lower intensity PA (Borremans et al. 2010; Einarsson et al. 2015; Foley & McCubbin, 2009; Stanish & Mozzochi, 2000).

PA levels in children and adolescents with ID also decline with age, with sedentary behaviour increasing (Phillips & Holland, 2011). This trend continues into adulthood, as adults with ID have been reported to participate in little or no PA (Finlayson et al. 2009; Hilgenkamp et al. 2012; Ptomney et al. 2017). Since children and adolescents with ID have a higher prevalence of ill-health than their TD peers, increasing PA could help reduce these health inequalities (Maiano, 2010). Furthermore, as childhood PA is a predictor of PA in adulthood, promoting active lifestyles at a young age will help promote long-term activity and health (Telama, 2009; Telama et al. 2005).

Increasing PA through interventions is one method to promote active lifestyles and reduce the risk of chronic disease (Biddle et al. 2015). Numerous systematic reviews in TD children and adolescents have investigated the effects of interventions on PA levels and health outcomes (Biddiss & Irwin, 2010; Janssen & LeBlanc, 2010; Metcalf et al. 2012; Van Sluijs et al. 2007). These studies have highlighted that increasing PA through interventions is complex, with many studies only reporting small changes in PA post-intervention. However, this evidence-base does not exist for children and adolescents with ID.

PA in children and adolescents with ID is a neglected area of research, resulting in a lack of knowledge on the design and implementation of effective interventions (Frey et al. 2008; Hinckson & Curtis, 2013). Previous systematic reviews and meta-analyses have investigated effects of PA interventions on health-related outcomes in children and adolescents with ID; however, interventions which aim to increase PA in children and adolescents with ID have not been systematically reviewed (Harris et al. 2015; Johnson, 2009). Therefore, there is no evidence to suggest which intervention components and designs are most effective in increasing PA in children and adolescents with ID.

This study aims to address these gaps in the literature by investigating: 1) How effective are interventions to increase PA in children and adolescents with ID?; 2) What components have been used in interventions to increase PA in children and adolescents with ID?

Method

This study was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Moher et al. 2009).

Search strategy

A literature search was conducted to identify relevant studies which included interventions to increase PA in children and adolescents with ID. Seven databases were searched [MEDLINE, EMBASE, Education Resources Information Center (ERIC), Cumulative Index to Nursing and Allied Health Literature (CINAHL), PsychINFO, Cochrane Central Register for Controlled Trials (CENTRAL), and International Standard Randomised Controlled Trial Number (ISRCTN) trials registry] up to and including July 2016. A systematic search strategy was developed based on truncated key terms relating to ID, PA, and interventions. This was limited to children and adolescents (0-18 years), English language, and humans (full search strategies are presented as a supplementary file). Reference lists of included studies and relevant identified systematic reviews were also hand searched.

Eligibility criteria

To be included in this review, studies had to: 1) Include a sample of children and adolescents with ID (<18 years); 2) include an intervention, and; 3) measure PA pre- and post-intervention. To ensure the included sample was representative of children and adolescents with ID, studies where <50% of sample were aged <18 years or <50% of sample had ID were excluded. ID was defined as limitations in intellectual functioning (intelligence quotient < 70) and adaptive behaviour, which onset in childhood (<18 years; Schalock et al. 2010).

Study selection

Once duplicates were removed, records were electronically imported into Covidence software for screening (www.covidence.org). Title and abstract screening and full text screening were independently conducted by two reviewers (AMMcG & LH), with disagreements discussed with a third reviewer (CAM). Reliability between reviewers (AMMcG & LH) for title and abstract screening and full text screening were calculated in SPSS (version 23; SPSS IBM, New York, NY, USA) using Cohen's kappa scores, demonstrating almost perfect agreement (κ=.98 and κ=.85, respectively; Landis & Koch, 1977).

Data extraction

A data extraction form was designed using Excel and piloted. Two reviewers (AMMcG & LH) independently conducted data extraction. Study details, participant characteristics, study characteristics, intervention characteristics, and study findings were extracted. Initially, seven studies were included at this phase, with the authors of three studies contacted to request additional information; these studies included PA as a secondary outcome but did not report PA post-intervention. One author provided the requested post-intervention PA data and therefore this study was included. However, the additional two studies were excluded at this point as the required data were not provided.

Assessment of methodological quality

Methodological quality was assessed using the Standard Quality Assessment Criteria for Evaluating Primary Research Papers from a Variety of Fields (Kmet et al. 2004). The quantitative checklist enables studies from various designs to be assessed. As the current review included various study designs, this tool was deemed most appropriate. This assessment tool includes a 14-item checklist, as described in Table 1. For each item, studies were scored with; yes=2, partial=1, no=0, or N/A where appropriate. The final quality assessment score was calculated as a percentage of relevant items attained, with any N/A scores excluded from this calculation. Therefore, the exclusion of "N/A" scores from these calculations provides comparable quality scores between studies, regardless of design. Two researchers (AMMcG & LH) independently conducted quality assessment, with discrepancies discussed with a third reviewer (CAM). Reliability between the two primary reviewers (AMMcG & LH) was substantial (κ=.72; Landis & Koch, 1977).

INSERT TABLE 1 HERE

Data synthesis

Effect sizes were calculated for multiple comparisons (Morris & DeShon, 2002). All effect sizes were based on change in PA [calculated from mean change, standard deviation (SD) change and sample size] pre-post comparisons. The primary analysis was the effect size for between group comparisons obtained from randomised trials; treatment versus control/comparator intervention (treatment pre-post effect size minus control/comparator pre-post effect size; Borenstein et al. 2009). Within group effect sizes were calculated for treatment and control groups separately and for quasi-experimental studies. Effect sizes were calculated as standardised mean difference (*d*) and were interpreted as small effect (*d* = 0.20), moderate effect (*d* = 0.50) and large effect (*d* = 0.80) with a positive *d* illustrating a favourable effect of the intervention on increasing PA (Cohen, 1988). An imputed correlation coefficient was calculated [based on the variance of pre-post and change in PA reported in the study by Shields et al. (2013)] for studies that did not report the SD of the change in PA (Higgins & Green, 2011).

Heterogeneity was assessed using the Cochrane's Q statistic, with a significance level of $p<.05$ indicating evidence of statistical heterogeneity. I^2 statistic was used to quantify the

degree of heterogeneity, with I^2 ≥50% indicating substantial heterogeneity (Higgins et al. 2003). A random effects model was used to compute effect sizes (DerSimonian & Laird, 1986). The meta-analysis was conducted using Comprehensive Meta-Analysis (Version 3.0 for Windows: Biostat, Englewood, Colorado, USA).

Results

Literature search

The database searches identified a total of 4447 records to be screened; the full search and screening results are presented in Figure 1. More than one reason for exclusion was provided for 10 studies, including conducting research not involving children and adolescents with ID and not measuring PA as an outcome. A hierarchy was decided, and not involving individuals with intellectual disabilities was chosen as the primary reason for exclusion. Five studies met the eligibility criteria and were included in this review.

INSERT FIGURE 1 HERE

Study characteristics

The primary aim of three studies was to examine the efficacy of interventions on increasing PA (Gephart & Loman, 2013; Hinckson et al. 2013; Ulrich et al. 2011). Two studies included PA as a secondary outcome (Ptomey et al. 2015; Shields et al. 2013). Three studies used accelerometers to measure PA outcomes, including time (minutes/day) spent in light, moderate, and vigorous PA, and counts per minute (CPM; Ptomey et al. 2015; Shields et al. 2013; Ulrich et al. 2011). Subjective measures were proxy-reported from parents/carers in the form of an interview (Gephart & Loman, 2013) and questionnaire (Hinckson et al. 2013). Three studies employed a randomised controlled trial (Ptomey et al. 2015; Ulrich et al. 2011; Shields et al. 2013) and two studies were uncontrolled quasi-experimental (Gephart & Loman, 2013; Hinckson et al. 2013). Intervention duration ranged from five days to four months. Follow-up measures were conducted in three studies at seven weeks/one year

(Ulrich et al. 2011) and 24 weeks (Hinckson et al. 2013; Shields et al. 2013) from baseline. An overview of study characteristics is illustrated in Table 2.

INSERT TABLE 2 HERE

Location and setting

Three studies were conducted in the USA (Gephart & Loman, 2013; Ptomey et al. 2015; Ulrich et al. 2011), one in New Zealand (Hinckson et al. 2013), and one in Australia (Shields et al. 2013). Two interventions were conducted in the community, specifically a gym (Shields et al. 2013) and summer camp (Ulrich et al. 2011). One study was conducted in two special needs schools (Hinckson et al. 2013) and one was conducted in a residential group home (Gephart & Loman, 2013). The study by Ptomney et al. (2015) was conducted in the participant's home, with the intervention delivered remotely via video communication (Facetime) on computer tablets.

Participant characteristics

A total of 223 participants with ID were enrolled across studies. The mean sample size of studies was 41 (range 17 to 61). Two studies included participants with mild to moderate ID (Ptomey et al. 2015; Shields et al. 2013); one included participants with mild, moderate, severe, and profound ID (Gephart & Loman, 2013); and two studies did not report the level of ID (Hinckson et al. 2013; Ulrich et al. 2011). Studies included individuals with Autism (Gephart & Loman, 2013; Hinckson et al. 2013; Ptomey et al. 2015) and Down syndrome (Hinckson et al. 2013; Ptomey et al. 2015; Shields et al. 2013; Ulrich et al. 2011). The mean age range of participants was 14 to 18 years. Two studies included participants over the age of 18 years, although adult participants represented <50% of the sample (Hinckson et al. 2013; Shields et al. 2013). More male participants were recruited across studies (58% male) than female participants. Weight status was heterogeneous and ranged from underweight to obese.

Interventions

Interventions were multi-component weight management programmes (Gephart & Loman, 2013; Hinckson et al. 2013; Ptomey et al. 2015), a single-component PA intervention (Shields et al. 2013), and a skill development training programme (Ulrich et al. 2011). A description of the intervention components is presented in Table 3. Only one study provided a theoretical framework as the foundation on which their intervention was based. Ulrich et al. (2011) founded their intervention on the principles of dynamic systems theory (Kamm et al. 1990). Interventions were based on evidence from clinical guidelines on weight management (Gephart & Loman, 2013; Ptomey et al. 2015), and PA (Shields et al. 2013), with one adapted from an existing weight management intervention in the general population (Hinckson et al. 2013).

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Multi-component weight management interventions

The multi-component weight management interventions were primarily based on a health education approach, which included providing dietary advice only (i.e. did not prescribe a specific energy intake to induce an energy deficit).

Intervention sessions included advice on portion sizes, healthy and unhealthy food groups, and food labelling. Only one study provided quantitative dietary advice in terms of an individualised daily energy deficit diet (Ptomey et al. 2015). All multi-component weight management interventions incorporated behavioural change components and a PA component. One study used a health education approach and advised participants to gradually increase their PA to a health-enhancing level (Hinckson et al. 2013). One study included active participation in PA, in the form of swimming and pool-based activities (Ptomey et al. 2015). Behaviour change was primarily focussed on goal setting, with additional techniques such as self-monitoring, performance feedback, and rewards incorporated across the interventions. Tablet computers were used in one study to help

facilitate behaviour change (Ptomey et al. 2015). These were used to monitor PA through data received from self-monitoring activity tracker devices (Fitbit) and monitoring of nutritional intake through the application (Lose it!). Moreover, the tablet computers were also used to deliver the intervention remotely through Facetime with the facilitator. The interventions were delivered by trained professionals including a registered nurse (Gephart & Loman, 2013) and registered dieticians (Hinckson et al. 2013; Ptomey et al. 2015). Additional support from a physiotherapist, teachers, senior management, and social workers was sought in the implementation of one weight management intervention (Hinckson et al. 2013).

PA intervention

The single-component PA programme developed by Shields et al. (2013) incorporated resistance/strength-based exercises. This intervention was based on American College of Sport Medicine (ACSM, 2013) guidelines and involved training of the major muscle groups. Prescribed exercises included seated chest press, knee extensions, and calf raises, which were performed using pin-loaded weight machines. Undergraduate physiotherapy students led the sessions on a one-to-one mentoring basis with the participants. Sessions were twice per week (45-60 minutes), with the weight increased once participants could perform three sets of 12 repetitions of each exercise.

Skill development training intervention

The skill development programme aimed to teach children and adolescents with ID to ride a two-wheeled bicycle (Ulrich et al. 2011). This was based on the dynamic systems theory of motor development (Kamm et al. 1990). Adapted bicycles (based on a two-wheeled bicycle with stabilising rollers replacing the back wheel), which were tailored to the specific riding skill level of the participants, were used to increase the confidence of participants' in developing a new skill without the fear of falling off the bicycle. Participants attended a 'summer camp' format, in which they attended the programme for 75 minutes on five consecutive days. Progression on to the next level of difficulty (eight levels of roller stabilisers) was assessed when participants could confidently ride for 10 minutes on the

adapted bicycle, and successful completion of the intervention was determined if participants could ride independently on a 2-wheel bicycle for a minimum of 9 meters.

Baseline PA levels

Light intensity PA was most prevalent, with participants in one study engaging in approximately 5 hours per day (Ptomey et al. 2015). MVPA ranged from one minute to 49 minutes per day (Ptomey et al. 2015; Ulrich et al. 2011). Gephart & Loman (2013) did not report PA intensity, however, 49% of participants did not engage in any PA, and only 16% engaged in ≥60 minutes per day. Two studies reported mean CPM, which ranged from 285 to 336 (Shields et al. 2013; Ulrich et al. 2011). Hinckson et al. (2013) reported PA type, which included physical education, walking, swimming, and active play. Participants spent the most time engaged in walking (2.96; SD 2.96 hours/week) and active play (2.9; SD 1.8 hours/week).

Methodological quality assessment

Quality assessment percentage scores are described in Table 2; full quality assessment is presented in a supplementary file. The mean score was 69.1% (SD 15.2%). The research objectives were demonstrated to be sufficiently described (Q1) and assessed by the appropriate study design (Q2) in all studies. The remaining questions were fully or partially fulfilled at least 50% of the time, with the exception of blinding of investigators (Q6) and participants (Q7). Only one randomised control trial (33%) successfully reported blinding of researchers conducting the study (Shields et al. 2013). In the two studies where blinding of the intervention to the participants was considered possible, neither report doing so (Ptomey et al. 2015; Shields et al. 2013). In general, studies recruited small sample sizes and did not provide sample size calculations or an appropriate justification for the sample size.

Attrition

In general, attrition was low (range = 1.5% – 15.3%; Ptomey et al. 2015; Shields et al. 2013; Ulrich et al. 2011). However, two studies did not report attrition, therefore it is uncertain whether or not participants withdrew from engaging in these studies (Gephart & Loman, 2013; Hinckson et al. 2013). Ulrich et al. (2011) recorded the highest attrition rate, due to the long waiting time of control participants before receiving the intervention.

Effect of the intervention on change in PA

Between-group intervention versus control/comparator effect

Effect sizes were computed for four of the studies included in this review (one study reported insufficient data, which was requested from the authors but not provided). The effect sizes for each PA outcome (i.e. light, moderate, vigorous PA, and CPM) are reported for each comparison (Table 3). Only two studies could be pooled in a meta-analysis and individual effect sizes were reported for pre-post comparisons for treatment and control/comparator groups and quasi-experimental studies separately. Meta-analyses were conducted on studies using objective measures and revealed that interventions were not more effective post-intervention (*d*: 0.20; 95% CI -0.57, 0.97; p=.61) in comparison to no treatment control (Table 4). However, there was a moderate effect of interventions at follow-up (*d*: 0.49; 95% CI 0.14, 0.84; p=.01). Full meta-analysis comparison results and forest plots for pre-post and follow-up are presented as supporting information. The moderate effect of treatment interventions, resulted from maintenance of (Shields et al. 2013) and an increase in PA (Ulrich et al. 2011) in comparison to a decrease in PA in the no treatment control intervention. One study compared the effect of two active weight management interventions (Ptomey et al. 2015) and found that different prescription methods in terms of energy deficit diets did increase total PA (*d*: -0.22; 95% CI -1.10, 0.66; p=.63).

INSERT TABLE 4 HERE

Within-group intervention effect

Within-group comparisons are reported as ancillary evidence to the aforementioned between-group comparisons from randomised controlled trials. In general, interventions had a small non-significant effect on PA post-intervention (*d* range -0.61 to 0.67). One study reported a moderate effect, approaching statistical significance (*d*: 0.57; 95% CI 0.01, 1.13; p=.05) for vigorous PA post-intervention (Ptomey et al. 2015). Two studies reported significant effects of the interventions on increasing PA at follow-up time points, which were not evident immediately on completion of the intervention. Hinckson et al. (2013) reported at six months follow-up participation in physical education was significantly increased (*d*: 0.67; 95% CI 0.16, 1.18; p = 0.01). Moreover, Ulrich et al. (2011) found a small significant effect of their skill development intervention in increasing MVPA at 12-month follow-up (*d*: 0.38; 95% CI 0.04, 0.27; p=0.03) and a moderate increase in average CPM (*d*: 0.52; 95% CI 0.18, 0.87; p<.01). Although statistically significant, the intervention effects reported were small in absolute terms, ranging from an increase of 3.6-9.5 minutes/day and 9.5-67.9 CPM.

Within group control effect

Participants in the no treatment control intervention did not increase PA (*d* range -0.43 to 0.03). PA levels were, in general, reported to decrease post-intervention and follow-up. Ulrich et al. (2011) reported that average CPM (*d*: -0.30; 95% CI -0.55, -0.04; p=.02) and MVPA (*d*: -0.43; 95% CI -0.69, -0.16; p<.01) decreased post intervention.

Discussion

This systematic review highlighted the paucity of interventions aiming to increase PA levels in children and adolescents with ID. The meta-analysis demonstrated that interventions were not effective at increasing PA levels post-intervention, compared to no treatment control. The pooled estimate at follow-up revealed a significant moderate effect of

treatment interventions in comparison to no treatment. However, this was primarily due to a decrease in PA in the control group.

Although this analysis was based on a small number of studies, the results demonstrate that the design of existing interventions are not effective for children and adolescents with ID, which may be due to a number of factors. Children and adolescents with ID have very set routines, therefore, interventions may require a longer period of time to enable habit development, which is a possible explanation for the increase reported within Shields et al. (2013). Specific to intervention delivery, little information was provided on the individuals facilitating the interventions and whether they were suitably trained to support behaviour change towards increased PA in children and adolescents with ID. Seasonal effects were also reported in the primary studies as contributing to the decline in PA levels, which may have contributed to the lack of change (Gephart & Loman, 2013; Hinckson et al. 2013; Ulrich et al. 2011). On the other hand, selective reporting of analysis to a specific subgroup of children and adolescents who learned to ride their bike may have exacerbated the effects and thus limit generalisation (Ulrich et al. 2011).

The generalisation of public health research conducted in the general population to individuals with ID is common practice, without clear evidence or justification. Interventions in the present review were developed based on existing interventions and theories from the general population. However, interventions involving TD children have, in general, not been effective in increasing PA levels, with only small increases reported; therefore, this is not a strong or effective evidence-base from which to draw from (Biddiss & Irwin, 2010; Janssen & LeBlanc, 2010; Metcalf et al. 2012; Van Sluijs et al. 2007). Furthermore, the applicability of behaviour change theories and techniques for increasing PA has been questioned in individuals with ID due the complexity and level of abstraction required, limited cognitive abilities, need for additional social support, and level of understanding of individuals with ID (Kuijken et al. 2016; Melville et al. 2015; Robertson et al. 2000; Willems et al. 2017).

Understanding the clinical effectiveness of interventions is also limited by a lack of population-specific research. Intervention effectiveness in the primary studies was based on a p-value of the significance of null hypothesis testing, which demonstrated some significant improvements in increasing PA at follow-up. Based on the recommendation of 60 minutes

activity per day, the actual changes reported were not sufficient for health benefits (Chief Medical Officers, 2011). However, research including children and adolescents with disabilities was not included in the development of these guidelines, and there is limited evidence on the relevance of these PA guidelines and existing dose-response relationships for children and adolescents with ID (Bull et al. 2010). Therefore, children and adolescents with ID may gain health benefits from shorter duration or less intense periods of PA. Conducting minimum clinical important difference (MCID) analysis is one potential method to examine any 'meaningful' results (Froehlich, 1998). Finally, the lack of population-specific measurement research in children and adolescents with ID limits the accuracy of results (McGarty et al. 2014). Therefore, valid and reliable methods, which are population-specific and sensitive to change, are essential to test the effect of interventions (McGarty et al. 2016).

In general, the lack of population-specific research in individuals with ID has shown to further exacerbate the health inequalities experienced by this group (Lorenc et al. 2013). Population-specific research and guidelines are therefore required to progress this area of research and develop effective interventions tailored specific to children and adolescents with ID. It is also important to note that interventions in adults with ID have not been effective in promoting significant or clinically meaningful increases in PA (Bergström et al. 2013; McDermott et al. 2012; Melville et al. 2015). Therefore, the lack of knowledge on the development of effective interventions is a wider issue in PA research in individuals with ID.

The meta-analysis was limited by the varied quality and reporting of studies in this review, thus raising questions over the internal validity of the findings. Results from quasi-experimental studies should be interpreted with caution due to increased risk of bias from unknown confounding factors and reverse causality. Despite this, the inclusion of quasi-experimental studies is important to address the present research aim due to the lack of randomised controlled trials conducted. This is in contrast to the abundance of evidence of interventions designed to increase PA in TD children and adolescents (Metcalf et al. 2012; Van Sluijs et al. 2007). Furthermore, studies reported small sample sizes which is typical of PA research conducted in children and adolescents with ID (Hinckson & Curtis, 2013). To improve study quality and inferences made, it is important that reasons for small sample sizes are discussed to add to the knowledgebase relating to recruitment strategies, response

rates, and compliance issues. Further, power calculations are required to establish how many participants are needed to answer study research questions (Jones et al. 2003).

Strengths and limitations of the review

A key strength of this review was the rigorous methodological design which adhered to PRISMA guidelines (Moher et al. 2009). Numerous databases were systematically searched using a comprehensive strategy to ensure all relevant research was identified. The quality of this review and reliability of findings is enhanced by double screening and reviewing, and the strict inclusion criteria applied ensured studies were relevant to the target population. Finally, to the authors' knowledge, this is the first meta-analysis to quantify the effectiveness of interventions to increase PA in children and adolescents with ID. However, publication bias could not be reliably assessed due to the limited number of included studies (Higgins & Green, 2011).

Recommendations for future research

The small number of studies identified and included limits the scope of recommendations on how best to intervene and increase PA levels amongst children and adolescents with ID. Thus, it is suggested that there is a need to fill the current gap in the literature and produce more high quality formative studies which provide valuable detail on the participants (i.e. level of ID), attrition rates, and compliance with PA measures. The generation of better theoretical and evidence-based studies is crucial to inform the development of future interventions. Researchers should also consider involving children and adolescents with ID and their parents in the development of studies to increase the relevance of interventions to participants' lives and needs.

Conclusion

In conclusion, increasing PA in children and adolescents with ID through intervention is an essential but under-researched area. Intervention studies have included varied designs and components, and differ in methodological quality. The meta-analysis found that interventions were ineffective at increasing PA levels in children and adolescents with ID. Therefore, recommendations on effective intervention components cannot be generated. Instead, there is a need for future research to form population-specific theoretical and

evidence-based interventions in order to effectively increase PA and reduce the health inequalities experienced by children and adolescents with ID.

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Table 1. Quality assessment checklist

Criteria		Yes (1)	Partial (1)	No (0)	N/A
1	Question / objective sufficiently described?				
2	Study design evident and appropriate?				
3	Method of subject/comparison group selection *or* source of information/input variables described and appropriate?				
4	Subject (and comparison group, if applicable) characteristics sufficiently described?				
5	If interventional and random allocation was possible, was it described?				
6	If interventional and blinding of investigators was possible, was it reported?				
7	If interventional and blinding of subjects was possible, was it reported?				
8	Outcome and (if applicable) exposure measure(s) well defined and robust to measurement / misclassification bias? Means of assessment reported?				
9	Sample size appropriate?				
10	Analytic methods described/justified and appropriate?				
11	Some estimate of variance is reported for the main results?				
12	Controlled for confounding?				
13	Results reported in sufficient detail?				
14	Conclusions supported by the results?				

* Adapted from Kmet, Lee, & Cook (2004)

Table 2. Overview of studies aiming to increase physical activity levels in children with intellectual disabilities.

Reference	Study design and location	Study population			Study duration (Follow-up)	Intervention	Physical activity outcomes	Attrition	Quality Assessment
			Intervention	Control/ Comparator					
Gephart et al. (2013)	Quasi-experimental uncontrolled USA	Sample size: Level of ID: Mild: Moderate: Severe: Profound: Age: (years) Gender: (Male/Female) Weight status: (BMI percentile)	40 5% 17.5% 40% 35% 16.9 (2.8) range 8-20 32/8 56.39 (34.58)	None	4 months	Multi-component weight management	Time spent engaged in physical activity – categorised into 15minute time intervals. Assessed by proxy report from interviews with carers	Enrolled n = 40 Completion rate not reported	45.5%
Hinckson et al. (2013)	Quasi-experimental uncontrolled New Zealand	Sample size: Level of ID: Age: (years) Gender: (Male/Female) Weight status: (kg/m^2)	17 Not reported 14.0 (4.0) 10/7 31.0 (8)	None	10 weeks (24 weeks)	Multi-component weight management	Time spent in physical education (hours) Time spent in MVPA (hours/week) Time spent sedentary (hours/week) Assessed by proxy report	Enrolled n = 72 Completed n = 17 Attrition or reasons for exclusion not reported	77.3%

							from parents completing questionnaire		
Ptomey et al. (2015)	Randomised controlled trial USA	Sample size: Level of ID: Mild: Moderate: Age: (years) Gender: (Male/Female) Weight status: (BMI percentile)	**eSLD** 10 12 (60%) 8 (40%) 15.9 (1.8) 5/5 92.7 (6.0)	**CD** 10 13.9 (2.2) 6/4 90.4 (7.0)	2 months	Multi-component weight management	Time spent in light and moderate to vigorous physical activity (minutes/day). Objective assessment from wearing accelerometer.	Enrolled n = 21 Completed n = 20 Attrition rate = 4.8%	71.4%
Shields et al. (2013)	RCT Australia	Sample size: Level of ID: Mild: Moderate: Age: (years) Gender: (Male/Female) Weight status:	**PRT** 34 19 (56% 9 (26%) 17.7 (2.4) 19/15 27.3 (3.8)	**Control** 34 15 (44%) 25 (74%) 18.2 (2.8) 19/15 27.2 (5.6)	10 weeks (24 weeks)	Physical activity	Physical activity counts per minute. Objective assessment from wearing accelerometer.	Enrolled n = 68 Completed n = 67 Attrition rate = 1.5%	85.7%

		(kg/m^2)							
Ulrich et al. (2011)	Randomised controlled trial (waiting list control) USA	Sample size: Level of ID: Age: (years) Gender: (Male/Female) Weight status: (kg/m^2)	**EXP-L** 19 Not reported 12.4 (3.3) 9/10 24.3 (3.5)	**Control** 27 12.0 (1.9) 11/16 23.0 (4.8)	5 days (7 weeks/ 1 year)	Skill development training	Time spent sedentary (minutes/day) MVPA (minutes/day) Counts per minute Objective assessment from wearing accelerometer.	Enrolled n = 22 Competed n = 61 Attrition rate = 15.3%	65.4%

Values represent mean (standard deviation)

eSLD: enhanced Stop Light Diet; CD: conventional diet; PRT: progressive resistance training; EXP-L: experimental group who learned to ride a 2-wheel bicycle

Table 3. Summary of intervention components and results

Reference	Intervention components	Intervention setting and sessions	Results	
			Baseline PA Means (SD)	**Change PA Effect size (*d*; 95% CI)**
Gephart et al. (2013)	**Multi-component weight management** Health education programme delivered to carers. **Diet:** Advice including portion sizes control and healthy options when eating out. Goal to increase fruit and vegetable intake to 5 portions per day (3 servings offered to participants) **Physical activity:** Goal to increase activity to 30 minutes per day and to decrease screen time viewing to less than 2 hours per day **Behaviour change:** Goal setting and providing information on consequences of behaviour in general	**Setting:** Community Group homes **Session Duration:** Not reported **Session Frequency:** Not reported **Session Facilitator:** Registered nurse	**Max time (min) n (%)** 15 48 (13) 30 43 (12) 45 33 (9) ≥60 59 (16) Values are N (no. of youth reported more than 7 days within the 10 group homes exercising at the indicted maximum time) and percentage of total youth reported on for the month.	**Max time (min) n (%)** 15 63 (19) 30 54 (16) 45 14 (4) ≥60 38 (11) Note: d not calculated result represent post data at 4 months
Hinckson et al. (2013)	**Multi-component weight management** **Diet:** Health education approach including portion sizes and food groups, food labelling, eating out. **Physical activity:** Health education sessions (participants and parents) and active session with participants only i.e. swimming/pool activates **Behaviour change:** Motivational skills - Triggers, goal setting and rewards	**Setting:** Special needs schools **Session Duration:** 2 hours **Session Frequency:** 2/week **Session Facilitator:** Paediatric physiotherapist and dietician with support from	**Hours/week** **Physical Education** 1.8 (1.1) **Walking** 3.0 (2.9) **Swimming** 0.9 (0.4) **Active play** 2.9 (1.8)	**Post intervention** **Physical Education** 0.08 (-0.50, 0.66) p = 0.78 **Walking** -0.09 (-0.60, 0.44) p = 0.75 **Follow Up** **Physical Education** 0.67 (0.16, 1.18) p = 0.01 **Walking**

		teachers, teacher aids, senior management and social work.		0.26 (-0.21, 0.73) p = 0.28 Sample size not reported for swimming or active play, *d* not calculated.
Ptomey et al. (2015)	**Multi-component weight management** **Diet:** **eSLD:** Portion controlled meals - consisting of two entrées and two shakes per day. **CD:** EDD of 500 to 700 kcal/day was prescribed. **Both:** Recommended to consume fruit and vegetable intake (5 servings/day) **Physical Activity:** Advised to participate in moderate intensity physical activity and to gradually accumulate a total of 60 minutes per day (gradual increase in 10 minutes per week) at least 5 days per week **Behaviour change:** Self-monitoring and feedback	**Session Duration:** Introduction 90 minutes diet orientation session Rest of sessions 30 minutes **Session Frequency**: 1/week **Session Facilitator:** Registered dietitian	**Minutes/day** **Light:** **eSLD:** 282.5 (96.0) **CD:** 307.0 (110.2) **Moderate:** **eSLD:** 13.0 (9.5) **CD:** 19.8 (14.8) **Vigorous:** **eSLD:** 1.3 (0.5) **CD:** 12.0 (16.4)	**Light:** **eSLD:** -0.19 (-0.71, 0.34) p = 0.48 **CD:** -0.61 (-1.18, -0.05) p = 0.33 **Moderate:** **eSLD:** 0.11 (-0.41, 0.63) p = 0.68 **CD:** -0.14 (-0.38, 0.67) p = 0.59 **Vigorous:** **eSLD:** 0.57 (0.01, 1.13) p = 0.05 **CD:** -0.29 (-0.82, 0.24) p = 0.28
Shields et al. (2013)	**Physical activity** Progressive resistance/strength based exercise regimen based on principles of ACSM (2009)	**Setting:** Community gym **Session Duration:** 45-60 minutes	**CPM:** **PRT:** 336.0 (68.0) **Control:** 300.0 (132.0)	**Post intervention** **PRT:** -0.12 (-0.41, 0.16) p = 0.39 **Control:** 0.03 (-0.19, 0.25) p = 0.78

	Mode: Upper body, lower body and core/trunk exercises. Performed 3 times (12 repetitions of each exercise). **Intensity:** 60-18% of one-repetition maximum	**Session Frequency:** 2/week **Session Facilitator:** Undergraduate physiotherapy student mentors		**Follow-up** **PRT:** 0.01 (-0.31, 0.34) p = 0.94 **Control** -0.30 (-0.67, 0.07) p = 0.11
Ulrich et al. (2011)	**Skill development training** Specially engineered adapted bicycles (tailored specifically to individual riding ability) are used to assist children with intellectual disabilities to learn to ride a two-wheeled bicycle. Training on bicycles is gradually progressed over the training period to try and achieve the goal of riding a two-wheeled bicycle.	**Setting:** Community **Session Duration:** 75 minutes **Session Frequency:** Continuous once day/5 days **Session Facilitator:** Trained professionals from Lose the Training Wheels organization	**Average CPM** **EXP-L:** 284.6 (124.8) **Control** 314.2 (154.0) **MVPA (minutes/day)** **EXP-L:** 39.2 (23.7) **Control** 46.9 (29.2)	**Average CPM** **Post intervention** **EXP-L:** 0.15 (-0.13, 0.43) p = 0.30 **Control:** -0.30 (-0.55, -0.04) p = 0.02 **Follow-up** **EXP-L:** 0.52 (0.18, 0.87) p <0.01 **Control:** -0.16 (-0.57, 0.25) p = 0.44 **MVPA** **Post intervention** **EXP-L:** -0.11 (-0.39, 0.17) p = 0.45 **Control:** -0.43 (-0.69, -0.16) p <0.01 **Follow-up** **EXP-L:** 0.38 (0.04, 0.72) p = 0.03 **Control:**

	-0.27 (-0.68 to 0.15) p = 0.20

eSLD: enhanced Stop Light Diet; CD: conventional diet; CPM: counts per minute; PRT: progressive resistance training; EXP-L: experimental group who learned to ride a 2-wheel bicycle

Table 4. Meta-analysis of between-group effects on physical activity outcomes

Study	Effect size				Heterogeneity		
	K	*d*	95% CI	p-value	Q	I^2	p-value
Post intervention							
Intervention vs control	2	0.20	-0.57, 0.97	0.61	4.78	79.09	0.029
Intervention vs comparator	1	-0.22	-1.10, 0.66	0.63	-	-	-
Follow up							
Intervention vs control	2	0.49	0.14, 0.84	0.01	0.73	0.00	0.39

K: number of studies; WMD: weighted mean difference; CI: confidence interval; Q: heterogeneity statistic for the model; I^2: index of heterogeneity beyond within-study sampling error.

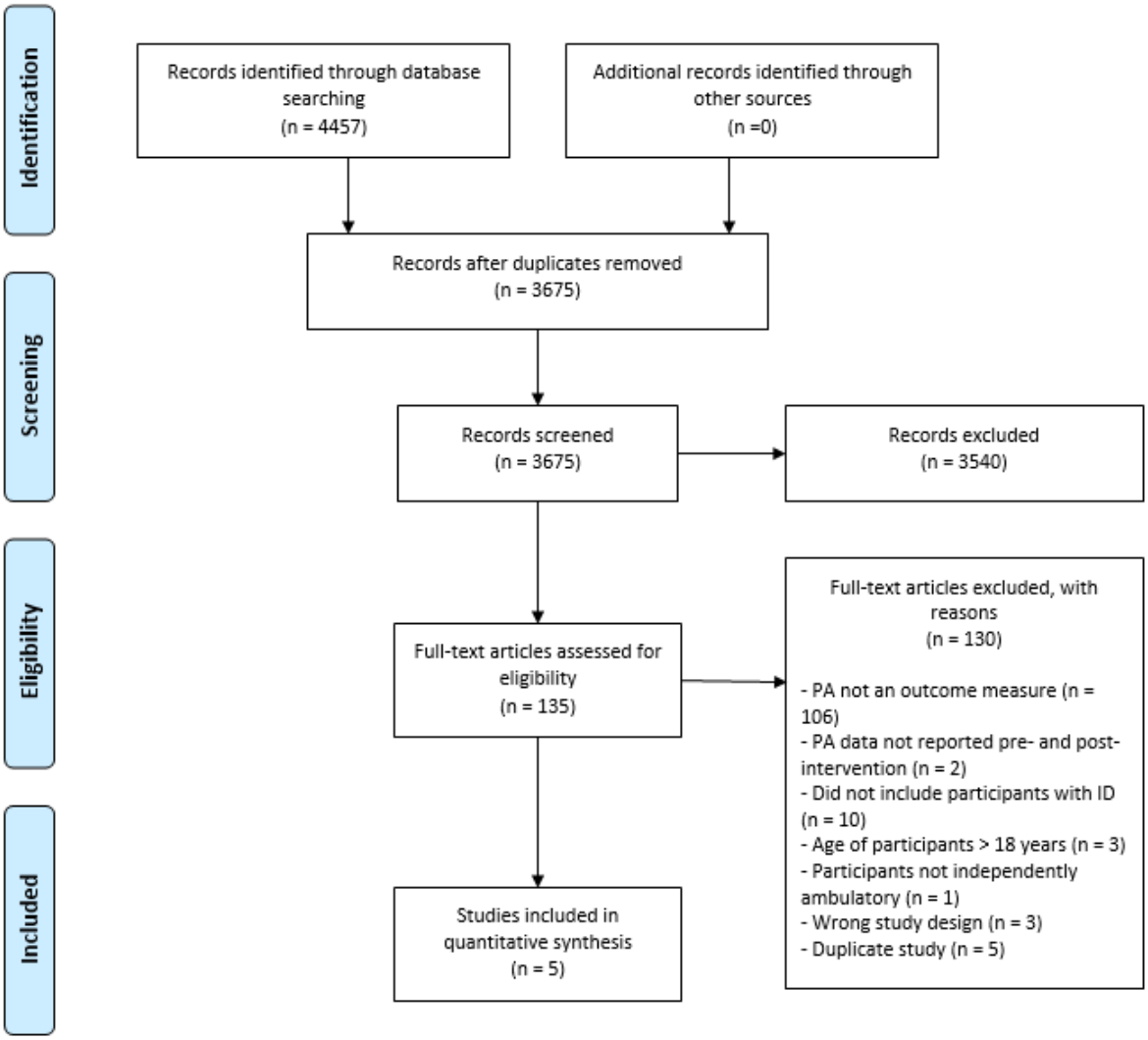

Figure 1. Search results and study selection flow chart. Adapted from Moher *et al.* (2009).