Interventions to promote physical activity for youth with intellectual disabilities

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Abstract
Objective. To describe interventions designed to promote physical activity for youth with intellectual disabilities. **Materials and methods.** A systematic review of nine databases until January 31, 2015 identified 213 citations. The inclusion criteria were: a) the study sample consisted of youth with intellectual disabilities, b) the study implemented an intervention to initiate, increase, or maintain physical activity, and c) quantitative or qualitative data were used to report the effectiveness of the intervention. Eleven articles from the 213 citations met this criterion. **Results.** Nine studies reported significant increases in physical activity behavior. **Conclusions.** Conclusions cannot be made regarding intervention components that impacted outcome variables, if the observed effects were specifically due to the intervention or if interventions could be maintained long-term. To advance the knowledge base in this area, a concerted effort should be made to increase rigor in study conceptualization and research design.

Keywords: physical activity; intellectual disability; youth; children; adolescents; intervention

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Intervenciones para promover la actividad física para jóvenes con discapacidad intelectual
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Resumen
Objetivo. Describir las intervenciones diseñadas para promover la actividad física para jóvenes con discapacidad intelectual. **Material y métodos.** Una revisión sistemática de nueve bases de datos hasta el 31 de enero de 2015 identificó 213 citas. Los criterios de inclusión fueron: a) la muestra del estudio consistió en jóvenes con discapacidad intelectual, b) el estudio implementado fue una intervención para iniciar, aumentar o mantener la actividad física y datos c) cuantitativos o cualitativos se utilizaron para informar la efectividad de la intervención. Once artículos de 213 citas cumplen este criterio. **Resultados.** Nueve estudios informaron aumentos significativos en el comportamiento de la actividad física. **Conclusión.** No se pueden establecer conclusiones con respecto a los componentes de intervención variables de resultado, considerando si los efectos observados fueron específicamente debido a la intervención o intervenciones podrían mantenerse a largo plazo. Para avanzar en la base de conocimientos en esta área, se necesita un esfuerzo concertado para aumentar el rigor en el estudio unívoco.

Palabras clave: actividad física; discapacidad intelectual; jóvenes; niños; adolescentes; intervención

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There is substantial evidence to support that regular participation in physical activity promotes physical and mental health benefits in children. The World Health Organization (WHO) recommends that children aged 5-17 years accumulate at least 60 minutes of moderate-to-vigorous intensity physical activity each day in order to improve cardiorespiratory and muscular fitness, bone health, and cardiovascular and metabolic health biomarkers.[1] Activities should be predominantly aerobic, but bone- and muscle-strengthening activities as well as vigorous physical activities should also be incorporated at least three times per week. Despite strong evidence to support the physiological and psychological health benefits associated with participation in physical activity, worldwide trends indicate that children fail to meet the recommended levels of physical activity.[1,2] This is equally true for children from Latin American; with the latest international 'Report Card' on children's overall physical activity reporting a 'C+' for Mexico and a 'D' for Columbia.[2]

The physical activity habits of children with intellectual disabilities have been studied far less thoroughly than those of typically developing children (i.e., children without disabilities), although research in this area has increased considerably over the past 5 or 6 years. In 2008, we published a review paper that aimed to summarize and critically analyze the existing literature on physical activity of youth with intellectual disabilities.[3] At that time, findings were variable with evidence to support that children with intellectual disabilities were less active, more active, and similarly active than their typically developing peers. We reported that significant methodological limitations and small sample sizes restricted the conclusions that could be drawn about the physical activity behavior of youth with intellectual disabilities. Overall, however, the research demonstrated that there was cause for concern. Empirical studies that have used motion sensors, such as accelerometers, to objectively measure physical activity have increased since that review was published. Consequently, the accuracy of results and the strength of the evidence have grown substantially.

International evidence suggests that children and youth with intellectual disabilities are less active than their peers without intellectual disabilities. A recently published study from Finland compared accelerometer-measured physical activity levels of children with intellectual disabilities and typically developing children aged 6-16 years.[4] Children with intellectual disabilities were found to be 40% less active than their peers, and none of the children with intellectual disabilities met the recommended guidelines for physical activity compared to 40% of typically developing children who did. A significantly lower proportion of children with intellectual disabilities used active commuting to school and fewer participated in organized sports. In the United States, Stanish and colleagues also found that physical activity levels measured by accelerometer were significantly lower in adolescents with intellectual disabilities than typically developing adolescents.[5] Only 6% of those with intellectual disabilities met the recommended time for moderate-to-vigorous intensity physical activity (MVPA) compared to 29% of their typically developing counterparts. Foley and colleagues compared the physical activity levels of young children with and without intellectual disabilities during the elementary school day and during out-of-school time. Accelerometer-measured physical activity levels were lower for children with intellectual disabilities compared to typically developing children during physical education class, recess, after school, and on weekends.[6] These results are corroborated by a study conducted in England that reported no children with intellectual disabilities met the physical activity recommendations, and participation in vigorous physical activity was essentially nonexistent.[7] Further, compared to published data on the general pediatric population in England, children with intellectual disabilities engaged in considerably more sedentary behavior. Shields and colleagues in Australia examined the physical activity levels of children with Down syndrome, a chromosomal abnormality involving intellectual disabilities, using accelerometers.[8] It was found that 42% of children with Down syndrome performed 60 minutes of MVPA each day which is lower than published reports of typically developing Australian children. Also, older children with Down syndrome accumulated significantly less physical activity compared to younger children. Though not without methodological shortcomings, these accelerometer studies demonstrate that overall youth with intellectual disabilities do not engage in sufficient physical activity to achieve health benefits.[9]

Some efforts have been undertaken to explain the low physical activity levels among children with intellectual disabilities. Given the intellectual, behavioral, social, and motor impairments that are often associated with an intellectual disability, it cannot be assumed that the factors that influence physical activity participation for these youth are the same as the general pediatric population. As such, studies of physical activity correlates among youth with intellectual disabilities have emerged. Research to date indicates that child and caregiver preferences for physical activity and caregiver educational level are positively related to physical activity participation among children with intellectual disabilities.[10-13] Barriers to participation have included lack of accessible programs, child's lack of interest, physical/motor challenges, behavioral difficulties, insufficient time, no location at which to participate, and transportation challenges. In addition, limitations in cognitive and adaptive functioning may warrant structure and supports in order for children with intellectual disabilities to participate successfully in activities. It is for these reasons that we must think uniquely when planning and implementing physical activity interventions for youth with intellectual disabilities.

The low levels of physical activity among children with intellectual disabilities coupled with their unique functional limitations and specialized needs, have created a significant need to develop and test interventions. The intervention research is growing and an evaluation of this work is fundamental to identifying effective strategies for increasing physical activity and health outcomes among children with intellectual disabilities. As such, the purpose of this review is to describe what characterizes interventions designed to promote physical activity for children with intellectual disabilities; and the effects of the interventions on overall physical activity levels and on health outcomes.

Materials and methods

Literature search strategy

A systematic search for physical activity intervention research in children and youth with intellectual disabilities was conducted as follows: (a EBSCOHost with selected databases: Academic Search Complete, CINAHL, Health Source: Nursing/Academic Edition, MEDLINE, PsychARTICLES, PsychINFO and SPORTDiscus and b) Scopus, a database that includes approximately 22 000 peer-reviewed journals from over 5 000 publishers until February 28, 2015. Since the terms physical activity, exercise, and fitness are often used interchangeably, albeit in error, all three were included for the outcome of interest and combined with population search terms (intellectual disability, mental retardation, Down syndrome, developmental disability, Prader-Willi syndrome) and (children, youth, adolescents). Reference lists of selected papers were also reviewed for relevant articles.

Study selection and data extraction

The following search criteria were established a-priori: a) the study sample was comprised of youth with intellectual disabilities aged 0-18 years, b) an intervention was used, c) physical activity was specified as dependent variable, and d) quantitative or qualitative data were used to analyse the efficacy of the intervention. All study designs were included to better capture the scope of the interventions being used in the field. Only original research papers published in indexed journals and that targeted youth with intellectual disabilities, not care givers, were included. Uncertainties about article inclusion were resolved through discussion and agreement between authors.

The systematic search process resulted in 951 citations. Of these, 738 were excluded as duplicates or because they did not meet the inclusion criteria based on abstracts or titles. Abstracts of the remaining 213 papers were examined and another 202 were excluded, with a final total of 11 papers included in the review (figure 1). Information on participant characteristics, study aims and design, study setting, intervention description, outcome measures and measurement time points, and physical activity results were extracted.

Results

Table I provides an overview of the few intervention studies focused on youth with intellectual disabilities that have included physical activity as a dependent variable. The scale of interventions ranged greatly from single subject/case study designs to randomized control trials. The number of randomized control trials was surprising since this type of research design is not common in exercise/physical activity research among people with disabilities.[14] Since the purpose of this review was to include studies using all types of research designs, randomized control trials were not assessed for quality using an objective tool such as the CONSORT checklist.[15] However, of the randomized control studies included in the current review, only one reported the random allocation process and who generated the allocation sequences.[16] O'Connor and colleagues[17] found that the quality of reporting for randomized control trial studies on engaging parents to increase activity in youth without disabilities was consistently poor and few met the CONSORT criteria. A tool such as the CONSORT checklist provides both a framework for reporting and process evaluation of randomized control trials, which improves the veracity and usefulness of the data. Five studies used accelerometry as an objective physical activity measure either alone or paired with a subjective measure (e.g. parent diaries). Youth physical activity is more intermittent and less predictable than adult physical activity, thus multiple measures may provide a more comprehensive picture of youth activity than single measures.[18] Two studies using accelerometry were conducted in infants with Down syndrome, but methodological research supporting the use of this instrumentation to assess physical activity in infants is

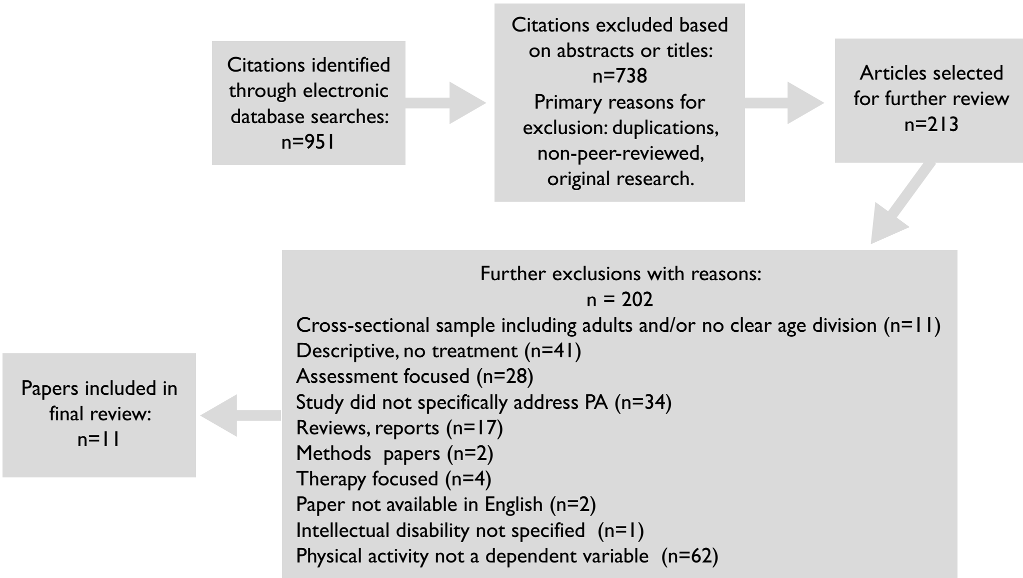

Figure 1. Article selection flow chart

lacking and requires further study.[19] There are also concerns that motion sensors may not capture movement differences in youth with intellectual disabilities and there are no intensity thresholds (i.e. cut-points) validated for this population segment.[20] Two accelerometry studies in this review used cut-points to assess physical activity intensity.[21,22] Ptomey and associates[22] reported using cut-points from the National Health and Nutrition Survey and referenced Troiano and colleagues.[23] However, Troiano and colleagues[23] used two different sets of cut-points for adults and children and the cut-points for children were adopted from research by Trost and colleagues.[24] Ptomey and associates[22] did not reference the Trost and colleagues[24] paper, thus it is unclear if the correct cut-points were used to distinguish between intensity levels. Unfortunately, the authors only reported *p*-values which limited the ability to interpret the data and findings. In contrast, Ulrich and associates[21]used the Actical accelerometer and cut-points developed for both the specific motion sensor model and children.[25] Ulrich and colleagues[21] also reported both *p*-values and mean±SD of physical activity variables, and this allowed the reader to better understand the data and findings. Poor consistency in reporting intervention methods and outcomes has also been observed in physical activity studies on youth without disabilities.[17,26]

Other methods of assessing physical activity included distance walked or snow-shoed[27] and parent proxy-reports.[28,29] Distance walked or snow-shoed has not been validated as a measure of physical activity and there is debate as to whether parent-proxy reports are a valid method of assessing youth physical activity.[30,31] Hinckson and Curtis[20] suggest that parent-proxy reports may be a suitable tool for assessing physical activity in youth with intellectual disabilities because of high parent-child contact during the day, but this conclusion was not based on research comparing child contact time between parents of youth with and without intellectual disabilities. As such, data based on proxy-reports of physical activity in youth with intellectual disabilities must be viewed with caution.

Discussion

None of the studies used direct observation to assess the impact of an intervention on physical activity, which is surprising since this method has frequently been used to assess physical activity levels in youth with intellectual disabilities.[20] Hinckson and Curtis[20] recommended that direct observation may be more suitable to assess physical activity in youth with intellectual disabilities than other objective tools because of the ability to capture short bouts of activity and changes in movement patterns. The best approach is to combine multiple measures, preferably at least one objective measure such as accelerometry, to best capture the unique aspects of physical activity in all youth,[18,32] including youth with intellectual disabilities.

Table I
Summary of study characteristics

	No. of studies (%)	*No. of participants (%) (Finished study)*
Year of publication		
2000 - 2009	3 (27.3)	
≥ 2010	8 (72.7)	
Country of publication		
USA	5 (45.5)	
New Zealand	2 (18.2)	
Taiwan	3 (27.3)	
Canada	1 (9.1)	
Number of participants		
Total number	5 (45.5)	Baseline: 252
Range (baseline)	1 (9.1)	End of study: 221
0-10	3(27.3)	(range 1 – 68)
11-20	2 (18.2)	
21-50		
51-100		
Mean age (years)		
Infancy 0-2	2 (18.2)	60 (27.2)
Early childhood 3-6	0 (0)	0 (0)
Later childhood 7-12	2 (18.2)	45 (20.4)
Adolescence 13-20…	7 (63.6)	114 (51.6)
Gender		
Male		122 (55.2)
Female		99 (44.8)
Physical activity measure		
Accelerometer + parent diaries	2 (18.2)	60 (27.2)
Accelerometer only	3 (18.2)	134 (60.6)
Gyroscope	1 (9.1)	2 (0.9)
Parent proxy-report	2 (18.2)	18 (8.1)
Distance	1 (9.1)	3 (1.3)
Intervention target participants		
Infants with Down syndrome	2 (18.2)	60 (27.1)
Youth with Down syndrome	2 (18.2)	114 (51.6)
Youth with intellectual disability and/or autism	1 (9.1)	3 (1.4)
Youth with intellectual disability, co-occurring with Down syndrome or autism	1 (9.1)	20 (9.0)
Youth with attending a segregated school (autism, Down syndrome, global developmental delay, intellectual disability)	1 (9.1)	17 (7.7)
Youth with mild-severe intellectual disability	4 (36.4)	7 (3.2)
Intervention setting		
Segregated school	4 (36.4)	23 (10.4)
Home	3 (18.2)	80 (36.2)
Community facilities (e.g. recreation centers)	3 (18.2)	115 (52.0)
Segregated school and community	1 (9.1)	3 (1.4)
Study design		
Randomized controlled trial	5 (45.5)	194 (87.8)
Pre-experimental design	2 (12.2)	18 (8.1)
Single subject design	4 (36.4)	9 (4.1)

There was a large participant age-range and the majority of studies included in the review involved adolescents. Several studies used a cross-section sample with the largest age-range between 7-20 years[29] representing a developmental span from late childhood to young adulthood. Physical activity differs according to developmental level[33] and none of the studies addressed physical activity from a developmental perspective or determined if participants met age-specific physical activity guidelines. The two studies involving infants were somewhat developmental in relating physical activity to walking onset[34,35] but no conclusions can be drawn about the impact of interventions on youth and intellectual disabilities meeting physical activity guidelines based on the studies in the current review.

Most interventions were conducted with students at segregated schools and this also reflects the geographical diversity of the studies. All of the studies at segregated schools were conducted outside the US,[27,29,36-38] while all but one of those conducted in the home or community were conducted in the US.[21,22,28,34,35] The US has mostly eliminated segregated schools for youth with disabilities in general, and this population segment is typically educated in the public school system.[39] Interestingly, no US school-based physical activity interventions for youth with intellectual disabilities were found in the literature search. There have been many multi-site, school-based interventions to address physical activity and health in youth without disabilities, and this approach is recommended to advance the field of physical activity and youth with intellectual disabilities.[14]

Effects on physical activity

Table II outlines the intervention delivery methods and outcomes on physical activity. Overall, 9 of the 11 studies (82%) studies reviewed reported that the intervention led to an increase in physical activity in the target population.[16,21,27-29,34-38] Ptomey and colleagues[22] did not observe an increase in physical activity, but there was a decrease in sedentary behavior and this could not be attributed to the intervention since there were no experimental and control group differences. Three studies came from the same laboratory and motivated

Table II
DESCRIPTION OF THE INTERVENTION STUDIES REVIEWED

Authors, country, aim & design	*Setting, sample & intervention description (including control group activities)*	*Outcome measures, measurement time points, and results*
Angulo-Barroso and colleagues[34]	Setting: home-based intervention	Outcome measures:
USA	Sample: infants with Down syndrome; N = 36 started, 30 completed intervention (12 females, 18 males), 26 completed 15-month follow-up measures.	PA assessment method: actiwatch on right ankle and iliac crest. Data were collected in 15 sec epochs. Parent activity logs.
Aim: comparison between high intensity and low intensity, weight-supported treadmill training on physical activity (PA)	Infants had to take at least six spontaneous steps in a minute, any time during a 5 min testing session. Age at study onset ranged from 5-12 months.	PA data was separated using 30% high-activity (Highact) and 70% low-activity (Lowact) criterion values.
Design: longitudinal, repeated measures, randomized comparative trial	Program name: no specific name	Other outcome measures: height, weight, head circumference, thigh skinfold, shank skinfold, umbilicus skinfold, motor ability using the Bayley Scales of Infant Motor Development.
	Intervention description: 16 infants were randomly assigned to high intensity treadmill training (HI) and 14 to low intensity treadmill training (LI).	Measurement time points: PA assessed over 24-hour period at study entry and every other month until independent walking (five data points per participant). 24-hour PA assessment occurred four times over 15 months following walking onset.
	Parents were instructed on how to implement the training at home and support the child on the treadmill.	
	HI group: treadmill speed, walking time and ankle weights were increased every two weeks in the HI group.	Intervention results: no significant group difference in age at onset of independent walking (HI group = 19.2±2.8 mos; LI group = 21.4±4.7 mos). The HI group spent less time in trunk and leg Lowact, and had a higher magnitude of leg Highact than the LI group.
	LI group: the protocol was not specified, but did not change during the intervention.	
	Duration: intervention duration continued five days/week until the infant could take three independent steps on the treadmill.	Post-intervention results: the HI group had more leg Highact, and less trunk and leg Lowact duration than the LI group.

A more detailed table and description of interventions in these articles is available at: http://go.iu.edu/1AO0 youth with intellectual disabilities to increase physical activity by receiving a preferred stimulus.[36-38] Different electronic configurations and modalities were used, but the most promising was a wireless gyration air mouse system.[36] The air mouse is a hand-held computer pointing device embedded with a micro-electro mechanical systems gyro sensor that allows control of a computer with hand movements. Using adapted software technology[40] the mouse could both detect precise limb movement and control a preferred stimulus (i.e. video projected on a flat screen TV). The air mouse was placed in a pants pocket and participants had to simply move at a certain intensity level in order to receive the stimulus. By controlling the stimulus through movement, participants were 80% more active than at baseline, although for a very short duration.[36]

Similar contingency-based interventions have been successfully used to increase physical activity in youth without intellectual disabilities,[41] as well the other studies included in this review involving youth intellectual disabilities.[37,38] These approaches used stationary bicycles[38,41] or other equipment[37] which may not generalize well to a practical setting due to high cost or large size. With the air mouse system participants merely needed to naturally move at a pre-determined intensity threshold to receive the preferred stimulus and this may be an optimal way to encourage physical activity, particularly in the home. These aforementioned studies are limited by small participant numbers and it is important to note that the same two youth participated in all three studies which may have habituated them to the stimuli. Regardless, this is an interesting approach to increasing physical activity in youth with intellectual disabilities that warrants further study.

The delivery of the other interventions varied considerably. Four were based on existing programs[21,22,28,29] and three cited evidence that the programs had been effectively used in other populations.[21,22,29] One used participating in a bicycle riding training camp,[21] two used infant treadmill training,[34,35] three used different types of exercise-based interventions including strength training,[16] walking and snow-shoeing with specific emphasis on self-monitoring, cuing, and reinforcement,[27] and a combination of strength/aerobic activities.[28,29] Two interventions targeted both physical activity and nutrition.[29,42] Most of the interventions incorporated a level of protocol individualization, even if a program was administered to a group.[16,21,27,28]

Several studies included parents by either having them help administer or facilitate the intervention[34,35,42] or by providing them with education materials and/or formal instruction.[28,29] There is an abundance of research on family based physical activity interventions for youth without disabilities, but the findings are generally inconclusive and do not support the efficacy of this approach.[17,26] While it is intuitive that parent contributions would be important to ensure the efficacy of a physical activity intervention for youth with intellectual disabilities, the existing research does not support this supposition. Three studies reported increases in physical activity[28,34,35] and two studies reported no improvements in physical activity.[22,29] The discrepancy in these findings can be attributed to many factors such as different outcome measures, subject populations, and intervention design. Schreiber and colleagues[28] measured physical activity using a single item question that asked the parent to circle the number of times their child exercised in the past two weeks on a scale of 1-15 and combined these data with qualitative information. The authors erroneously reported positive findings about the effectiveness of the intervention based on the parent's perception of the child walking "longer and faster" and the single item question. In addition, the other two studies that reported increased physical activity were based on infants with Down syndrome and parents implemented the treadmill intervention at home. Parents had control over the activity, which is a very different approach than parents influencing physical activity in other ways (e.g. verbal prompts, providing opportunities, etc.). As such, the studies reviewed indicate that parents either have no or limited influence on the physical activity of youth with intellectual disabilities.

Several studies examined the maintenance of intervention impact on physical activity at time points ranging from 3-15 months post intervention.[16,21,29,34] Of these studies, three reported that participants continued to exhibit elevated levels of physical activity 3,[16] 15[34] and 12[21] months post-intervention. Interestingly, Shields and colleagues[16] observed physical activity differences between experimental and control groups at three months, but not immediately after a progressive strength training intervention. This finding was attributed to the experimental group maintaining and the control group decreasing physical activity levels. Since there were no group differences in physical activity immediately post-intervention, it is incorrect to conclude that the intervention had an impact on physical activity levels in the target population.

Effects on other outcomes

All of the papers reviewed assessed a variety of outcome measures additional to physical activity, including selected anthropometrics (e.g. BMI, waist circumference),[16,21,22,29] eating behavior,[22,29] technology use,[22] walking onset,[34,35] and other physical fitness[16,21,28,29] or motor variables.[35] Hinckson and colleagues[29] examined physical activity, dietary habits, and overall health in youth with intellectual disability or autism. Several different outcome variables were included such as BMI, waist circumference, physical fitness (six-minute walk-test), sedentary behavior (screen time), sport (walking, swimming, active play) and physical activity (physical education). The impact of the intervention on these variables ranged from "unclear" to "very likely negative" and the only "possibly positive" effect was reported for the six-min walk 24-weeks post-intervention. Ptomey and colleagues[22] assessed anthropomorphic variables, several different aspects of energy/macronutrient intake, and diet quality and the intervention had a significant impact on select diet variables, but not anthropomorphic variables or physical activity. Ulrich and colleages[21] found that participation in a bicycle riding camp significantly improved physical activity, BMI and percent body fat in youth with Down syndrome, but not measures of leg strength and balance.

Inclusion of multiple and potentially correlated variables in an intervention with small subject numbers, is generally an ill-advised approach. Hadley and colleagues[43] reported that child obesity prevention and treatment programs that focused on one outcome (i.e. either nutrition or physical activity or weight loss) were more successful than those that tried to simultaneously focus on multiple outcomes. None of the studies reported power/sample size calculations or analysed dependent variable collinearity, and only one study reported post-analysis effect sizes.[21]

Conclusions

It is well documented that youth with intellectual disabilities are less active than peers without disabilities,[3] however there are few published efforts to improve physical activity in this population segment. This review illustrates that both the quantity and quality of physical activity interventions for youth with intellectual disabilities are lacking. Several interventions showed promising results and evidence of some benefits,[21,27,34-38] but reporting and design flaws make it difficult to generalize or replicate findings. Four of the single subject design interventions used a withdrawal component to verify intervention effects and were successful in improving physical activity.[27,36-38] However, none of the intervention approaches were based on behavior change theory or specifically targeted identified physical activity determinants which are important components of successful intervention design.[10,11,17] As a result, conclusions cannot be made regarding intervention components that impacted outcome variables, if the observed effects were specifically due to the intervention or if interventions could be maintained long-term.

To advance the knowledge base in this area, there needs to be a concerted effort to increase rigor in study conceptualization and research design. Physical activity intervention research design should be based on established behavior theory, particularly conceptualization of the problem and defining the target behavior that will be modified.[17,44] An increase in the number of randomized control trials is needed,[14] but studies using this design must address quality control in design and reporting to ensure usability of findings.[15,17] Finally, there are many challenges to conducting large-scale interventions in youth with intellectual disabilities, so researchers are encouraged to develop multi-site collaborative projects to increase sample size, strengthen research design and improve generalizability of findings.[14]

Declaration of conflict of interests. The authors declare that they have no conflict of interests.

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