RESEARCH ARTICLE

Meta-analysis of effectiveness of community-based interventions in improving physical activity in youth with intellectual disability

Anton Miguel M. Jordan^{1,2}, Frances Eunice Arcenal¹, Marie Stella Louise A. Perreras¹, Andreana Raina Rosa T. Crucillo¹, Rainier F. Canalita¹, Mary Rose S. Balce¹, Maria Eliza R. Aguila^{1*}

*Corresponding author's email address: mraguila1@up.edu.ph

¹Department of Physical Therapy, University of the Philippines, Manila, Philippines ²Department of Rehabilitation Medicine, Philippine General Hospital, Manila, Philippines

ABSTRACT

Background and Objectives: The effects of home or community-based interventions in increasing physical activity in youth with intellectual disabilities are yet to be known. Such information is important in designing the care for this population because of their increased susceptibility to lifestyle diseases that affect their quality of life. This study aimed to determine the effectiveness of home or community-based interventions in increasing physical activity levels in youth with intellectual disabilities.

Methodology: Eight databases were searched to identify intervention studies available in English that investigated the effects of home or community-based interventions on physical activity levels in youth with intellectual disabilities. Data were analyzed using descriptive statistics and meta-analysis using a random effects model. Effect sizes were calculated as standardized mean differences with 95% confidence intervals.

Results: Six randomized controlled trials were included in this review. Home or community-based interventions had a small effect in improving physical activity levels in youth with intellectual disabilities. These effects did not reach statistical significance for physical activity levels (d = 0.33; CI = -0.11 to 0.78; p = 0.14), number of steps per unit of time (d = 0.23; CI = -0.23 to 0.68; p = 0.32), and time spent inactive (d = 0.15; CI = -0.29 to 0.60; p = 0.50).

Conclusion: Home or community-based interventions may be effective in increasing physical activity levels in youth with intellectual disabilities. Further research with homogeneous outcome measures and larger sample sizes need to be conducted to build on the results of this review.

Keywords: Intellectual disability, physical activity, child, adolescent

Introduction

Regular engagement in physical activity (PA) is imperative for youth with intellectual disabilities (ID), regardless of associated medical diagnosis, because youth with ID experience greater health disparities than their typically developing peers [1]. Besides the increased susceptibility to chronic and lifestyle diseases, intellectual disabilities alone cause a "cycle of deconditioning" [2] in which the physical function and mobility of individuals deteriorate further with age [2]. Sedentary lifestyle and significant levels of inactivity have been identified to exacerbate these health problems [3]; and yet, this inactive behavior remains pandemic among youth with ID. A study conducted in Taiwan revealed that only 8% of youth with ID follow the physical activity guidelines of exercising at least three times per week for 30 minutes [4]. Likewise, a study from England showed that only 8% of youth with ID exhibited the characteristics of being physically active [5].

Furthermore, attempts to attain a more active lifestyle in this population are being hampered by inadequate access to quality health services, lack of information on appropriate health promotion strategies, and low socioeconomic status of the affected families [6–8]. In fact, several studies have shown that persons with ID have the highest poverty rates of all population groups [8] on top of having a lifetime economic cost of over one million dollars per person that is associated with their disability [9]. Their financial constraints, hence, make access to medical care, more so wellness initiatives, more challenging. Regular engagement in physical activities has been proven to have positive effects on typically developing youth in terms of overall fitness, motor skills, psychological health, social participation, and behaviors [10]. The participation of youth with ID in PA is therefore considered a health priority as it optimizes function, increases independence, and enhances overall quality of life.

Intellectual disabilities are long-lasting, with persistent consequences to watch out for. As such, the expenses of care for individuals with ID may be significant [7]. Cost-effective health interventions are, therefore, vital because of the prospective need for long-term care. Home and community-based interventions provide cost-effective options for treatment by eliminating transportation expenditures, utilizing available equipment and resources, and eliminating the need for consistent specialized supervision contrary to what institutional care offers [11]. These interventions cut down the miscellaneous expenses that may potentially bar families to receive quality health care [12].

A recent review and meta-analysis by McGarty and colleagues investigated the effectiveness of physical activity programs in different settings for children and adolescents with ID [13]. Findings suggest that these interventions are ineffective, possibly partly due to a limited number of studies with small sample sizes. Thus, it was concluded that this area of study is essential but under-researched. This present review, therefore, supplements current knowledge on effective interventions for youth with ID while focusing on frugal interventions. The aim of this systematic review is to determine the effectiveness of home or community-based interventions in increasing PA levels in youth with ID. Such interventions would be consistent with the growing need for frugal alternatives. More importantly, determining the effectiveness of home or community-based interventions would help improve the overall functioning and quality of life of youth with ID. Clinicians, families, and other health professionals concerned with exercise provision, and policymakers intent on addressing the health needs of youth with ID would gain insight for clinical and policy decisions.

Methodology

Protocol and Registration

The protocol for this systematic review was registered with PROSPERO (registration number CRD42017080687). This review followed methodological and reporting guidelines from the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Statement (PRISMA) [14].

Eligibility Criteria

Studies were included if they were intervention studies (randomized controlled trials, clinical trials, or cohort studies) that investigated the effects of home or community interventions on PA levels in youth with ID, regardless of associated medical diagnosis. Physical activity levels should be one of the outcomes. Studies were also included if they were in English with available full text. No restrictions were set on the nature of comparison interventions and whether interventions were movement-based or non-movementbased. Youth was defined as any age less than 18 years old, which would include children or adolescents. Home or community-based interventions was defined as any interventions done in either home or community setting.

Information Sources

The following databases were searched for relevant studies from their inception to November 2017 for relevant studies: Cochrane Library, Cumulative Index to Nursing and Allied Health Literature (CINAHL), Excerpta Medica DataBASE (EMBASE), Medical Literature Analysis and Retrieval System Online (MEDLINE), Physiotherapy Evidence Database (PEDro), PsycINFO, The Allied and Complementary Medicine Database (AMED), and Web of Science.

Search Strategy

We used a search strategy using a combination of Medical Subject Headings (MeSH) with the following keywords: "intellectual disability" OR "developmental disability" OR "learning disability"; "youth" OR "adolescent" OR "child" OR "pediatric"; "communit*" OR "home*" and related terms; "physical activity" OR "physical fitness" and related terms. This search strategy was translated for use in all other databases listed. The search was limited to English and human studies. In addition, researchers scanned the "related articles" link of databases and hand-searched reference lists of included studies and relevant identified systematic reviews. The full details on the search strategy for Medline are provided (see Appendix A). The search began in November 2017 until December 2017. An updated search was done in December 2021 but no additional studies were included.

Study Selection

Duplicate citations were removed by the reviewers using EndNoteTM X7.3 (Thomson Reuters. Endnote. New York:

Thomson Reuters; 2015). Pairs of reviewers (AJ and MB, FA and MP, RC and AC) then independently determined the eligibility of the remaining citations by applying the set inclusion criteria through title and abstract screening. Disagreements between the pairs were resolved by a third reviewer (MA).

Quality Assessment

Each included study was critically appraised for its methodological quality using a 27-item checklist, as described in Appendix B. Items 1-26 were from the Checklist for Measuring Study Quality, which is a commonly used tool to assess any study describing health care interventions [15]. The last item regarding power (item 27) was modified to answer whether it was reported or not.

Data Extraction

A data extraction form was designed and piloted by all reviewers. Revisions were then made to include other pertinent details. Data were extracted and cross-checked by three pairs of reviewers until a consensus was reached. Disagreements between the pairs were resolved by a third reviewer (MA). Extracted data included study characteristics (research design, sample size, intervention and comparison characteristics, length of program, measurement tools, and main results) and participant characteristics (age, diagnosis).

Data Analysis

Age, total number of participants, and sex were analyzed using descriptive statistics. The primary outcome assessed in this review was PA levels. Meta-analysis was done by pooling the results of studies based on standardized mean differences with a 95% confidence interval using the Comprehensive Meta-Analysis Software (Version 3 for Windows; Biostat, Englewood, CO, USA). Random effects model was applied in consideration of possible heterogeneity of the studies. Standardized mean differences were used as the effect size for all continuous outcomes with 0.20 interpreted as small, 0.50 medium, and 0.80 or higher as large effect size following Cohen's criteria [12]. A p value of 0.05 or lower was considered statistically significant. Visual inspection of the forest plot and computation of I2 statistic were done to determine statistical heterogeneity. I2 value of 50% or higher indicated significant statistical heterogeneity [16]. Subgroup analyses were conducted to further investigate any heterogeneity on outcome measures that may be influencing results. When data were available, number of steps were analyzed using means and standard deviations. Similarly, available data on time spent inactive were analyzed using

means and standard deviations of percentage of time and actual minutes. All outcome variables of PA which used objective measures were included in the meta-analyses. For all outcome variables, the relevant data were the postintervention values at 24 weeks. If data for this timeframe were not reported, values closest to 24 weeks were used in the analyses.

Results

The database searches yielded a total of 554 records. From these, a total of six studies were included in the review, four from full-text screening and two from handsearching. The flow of the studies, including the reason for exclusion and screening for eligibility, are shown in Figure 1.

Study Characteristics

All six included studies were randomized controlled trials. The methodological quality varied minimally from fair to good (see Appendix B). Four out of six studies [11, 18-20] were rated good, scoring > 70%, and the other two studies [17,18] were rated fair, scoring > 50%. All included studies scored 71% on internal validity. For external validity, one study [19] scored 100% while the rest scored 30-70%.

Four out of six studies [11, 18, 21, 22] had interventions set in the community, while two studies [19, 20] were set at home. The interventions included progressive resistance training [11, 18], skills development bicycle training [22], internet-based training program [20], and multicomponent physical activity programs [19,21]. Table 1 presents an overview of the study characteristics and their key results.

Home or community-based interventions were compared to regular school program which included gymnastic lessons [21], or recreational activities [11,18], or to usual activities or care or physical therapy [11,19,20], or to no intervention [22].

All six studies measured PA using objective measures. Five out of six used accelerometers [11,18,20,22] to measure the number of steps per unit of time as well as the time spent inactive. One study computed the ratio of total energy expenditure (TEE) to sleeping metabolic rate (SMR) or resting metabolic rate (RMR) [21]. Two out of six studies measured PA using subjective measures including the Child-Adapted Activity Questionnaire for Adults and Adolescent (AQuAA) [19] and the Multimedia Activity Recall for Children and Adolescents (MARCA) [20].

Table 1. Summary of Included Studies

Reference	Study Design	Intervention	Comparison	Instrument	Length of the Program	PA Outcomes	Results
Bania <i>et al.</i> (2016)	RCT	Progressive resistance training (PRT) in community gymnasium	Usual care (recreation and physiotherapy)	Activity Monitor (ActivPAL^TM)	12-week lower limb progressive resistance training	Steps/day Time spent in sitting and lying down	Participating in a 12-week progressive resistance training program does not increase the daily physical activity of adolescents and young adults with bilateral spastic CP and mild-to- moderate walking disabilities.
Shields <i>et al.</i> (2013)	RCT	PRT in community gymnasium	Social program and recreational activities	Rt3 activity monitor (Stayhealthy Inc., Monrovia, CA)	10-week progressive resistance training (PRT) programme	Count/min	Those who participated in PRT maintained their physical activity levels at 6-months compared to a corresponding decrease in the control group.
Ulrich <i>et al.</i> (2011)	RCT	Bicycle training	No intervention	Actical accelerometer s (Philips Respironics Inc., Bend, Oregon)	5-day bicycle training	Time spent in sedentary Time spent in moderate to vigorous activity Count/min	The experimental group reduced the average amount of time spent in sedentary activities per day by 75 minutes.
Van Wely <i>et</i> <i>al.</i> (2014)	RCT	Counselling Home-based physiotherap y Fitness training	Usual physiotherapy	Ankle-worn bi- axial accelerometer Child-adapted Activity Questionnaire for Adults and Adolescent (AQuAA)	6-month physical activity stimulation program	Accelerometer 1. Number of strides per day 2. Time spent at medium-to- high stride rate 3. Time spent at high stride rate 4. Time spent inactive AQuAA (measured in METs) 1. Weekly time spent at moderate-to-vigorous intensity (>5 METs), 2. Whether children met the physical activity guideline (one hour daily at >5 METs) 3. Weekly time spent inactive (<2 METs)	There was no significant effect of the intervention on physical activity.
Maher <i>et al.</i> (2010)	RCT	Get Set (internet- based program)	Usual therapy care and activities	NL-1000 pedometer and accelerometer (New Lifestyles Inc., MO, USA) Multimedia Activity Recall for Children and Adolescents (MARCA)"	8-week internet- based lifestyle program / GetSet modules	Step counts Distance travelled MARCA score MVPA counts	At 10 weeks the intervention group showed no increased physical activity compared with the comparison group (weekly steps, weekly moderate- to-vigorous physical activity, weekly distance walked). There were no statistically significant differences for these outcomes at 20 weeks, or in self-reported physical activity at 10 or 20 weeks.
Van Den Berg-Emons <i>et al.</i> (1998)	RCT	45-min exercise sessions 4x/week School program	No extra physical training School program	Level of daily PA: ratio of total energy expenditure (TEE) to sleeping metabolic rate (SMR) or TEE to resting metabolic rate (RMR)	9-month training programme	Ratio of total energy expenditure to sleeping metabolic rate or resting metabolic rate (TEE:SMR or RMR)	Although training has only a limited effect on restoring the deficit in PA in children with CP, regular physical exercise is important in young children with spastic CP because it may prevent deterioration in body composition.



Table 2. Participant Characteristics

Reference	Number		Age (y) Mean (SD)		Gender (Male)			Gender (Female)			Diagnosis		
	Total	Ехр	Con	Total (%)	Ехр	Con	Total (%)	Exp (%)	Con (%)	Total (%)	Exp (%)	Con (%)	
Bania <i>et al.</i> (2016)	36	15	21	18.7 (2.3)	18.2 (1.7)	18.9 (2.7)	18 (50)	8 (53)	10 (47)	18 (50)	7 (47)	11 (53)	Cerebral palsy (GMFCS I-III)
Shields <i>et al.</i> (2014)	68	34	34	17.9 (2.6)	17.7 (2.4)	18.2 (2.8)	38 (55)	19 (56)	19 (56)	30 (44)	15 (44)	15 (44)	Down syndrome
Ulrich <i>et al.</i> (2011)	46	19	27	-	12.4 (2.2)	12 (1.9)	20 (43)	9 (47)	11 (40)	26 (56)	10 (52)	16 (59)	Down syndrome
Van Wely <i>et al.</i> (2014)	49	25	24	-	9.5 (1.5)	10 (1.8)	28 (57)	12 (48)	16 (66)	21 (43)	13 (52)	8 (34)	Cerebral palsy (GMFCS I-III)
Maher <i>et al.</i> (2010)	41	20	21	13.56	13.7	14.1	26 (63)	12 (60)	14 (66)	15 (37)	8 (40)	7 (34)	Cerebral palsy (GMFCS I-III)
Van Den Berg- Emons <i>et al.</i> (1998)	20	10	10	-	9.5 (1.6)	8.8 (1.1)	11 (55)	4 (40)	7 (70)	9 (45)	6 (60)	3 (30)	Cerebral palsy



PRISMA 2009 Flow Diagram

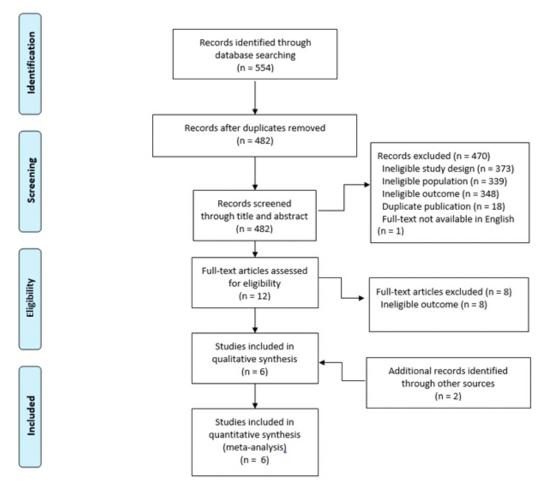


Figure 1. Flow of studies in the systematic review.

Participant Characteristics

A total of 256 participants were obtained across the studies for the outcome of PA levels. The mean sample size of the studies was 45 (range 20 to 68). The mean age of the participants was 18 (range 14 to 20). More male participants were recruited across the studies (53%). Four studies had participants with cerebral palsy [18,19,21], whereas two studies had participants with Down Syndrome [11,22]. One study specified their participants as having mild and moderate intellectual disability [11]. Table 2 summarizes the participant characteristics.

Effect of Intervention on Physical Activity

Home or community-based interventions had a small effect in improving PA levels in youth with ID that did not reach statistical significance (d = 0.33, CI: -0.11 to 0.78, p = 0.14; Figure 2). Moreover, there was evidence of significant heterogeneity among the included studies (I2 = 66.16%).

Sub-group analyses of physical activity measures revealed that home or community-based interventions demonstrated a small effect in improving number of steps per unit of time (d = -0.23, CI: -0.23 to 0.68, p = 0.33; Figure 3) as well as for time spent inactive (d = 0.15, CI: -0.29 to 0.60, p = 0.50; Figure 4) among youth with ID. These effects did not reach statistical significance.

Discussion

This study provides evidence that home or communitybased interventions have a small effect in improving PA levels in youth with ID that did not reach statistical significance. Similar results were obtained when analyzed further according to the measure of physical activity, namely time spent inactive and number of steps per unit of time. These results were obtained from six studies that fulfilled the eligibility criteria for this review and that have heterogeneous outcome measures and small sample sizes.

In the absence of other evidence on frugal interventions for youth with intellectual disability, results support the potential usefulness of home or community-based interventions in improving the physical activity of this population. The effectiveness of home or community-based interventions may be rooted in the following reasons. First, majority of the interventions did not require specialized supervision or equipment which may have made it feasible for the study participants to replicate the exercises even after the intervention period [11]. Second, the nature of the home- or community-based care fosters familiarity and comfort [23]. Such set-up may have allowed the child to adapt to the environment more quickly without being constrained by other external factors such as apprehension and poor socialization. Third, home or community-based exercises provide opportunities for self-monitoring of improvements over time which could be an effective tool for behavioral change [24].

These results build on the results of an earlier review by McGarty and colleagues which analyzed the effectiveness of PA programs in varied settings for children and adolescents with ID [13]. In the earlier review, varied physical activity programs showed a moderate effect in improving PA levels that did not reach statistical significance. This present review partially addresses issues identified in the earlier review, that is the need for evidence for frugal interventions, and echoes the call for more studies to be conducted in this area.

The pooled effects reported in this review may have been influenced by the results of the study of Van Wely and colleagues which favored the comparison group, visible on inspection of the forest plot (Figure 2) [19]. The different trend in this particular study is difficult to explain given that there is no apparent methodological or clinical difference between this study and the other studies included in the review.

To our knowledge, this is the first systematic review and meta-analysis that summarized evidence on the effectiveness of home or community-based interventions in improving PA levels in youth with ID. Families struggling with the burden of increased healthcare expenses associated with their children's long-term care would benefit from the costeffective interventions provided in this review. Healthcare professionals and policymakers may also gain insight from the results of this review, which would guide them in creating cost-effective programs for improving PA in the youth with ID.

However, results should be interpreted considering the limited number of studies included in this review, the small samples selected through convenience sampling, and heterogeneity in characteristics of participants, comparison interventions, and outcome measures. The setting of the studies being in generally high-income countries also limits the generalizability of the results for youth with ID who may not have the same access to quality health care. Further research with larger sample sizes, homogenous outcome measures with a consistent follow-up period, and in varied settings to represent variations in access and quality of



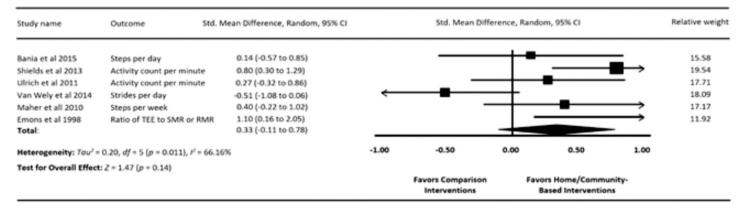


Figure 2. Forest plot showing effects of home or community-based interventions on physical activity levels.

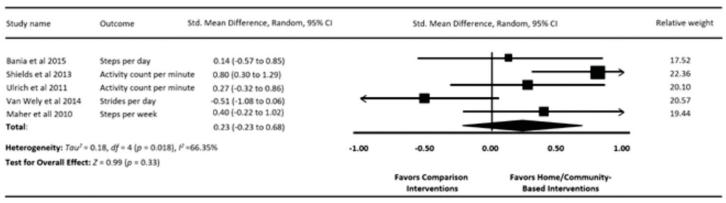


Figure 3. Forest plot showing effects of home or community-based interventions on number of steps per unit of time.

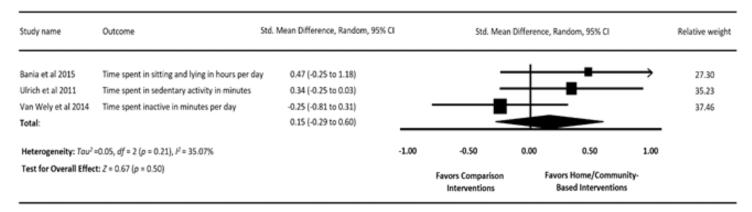


Figure 4. Forest plot showing effects of home or community-based interventions on time spent inactive.

health care is warranted. Moreover, future reviews are recommended to analyze subjective measures apart from objective findings.

Conclusion

This review provides initial evidence that home or community-based interventions may be effective in improving PA levels of youth with ID. Given the limited studies included in this review, small sample sizes, and varied comparison interventions and outcome measures, the effectiveness of these interventions remains inconclusive. Therefore, more studies with larger sample sizes and homogenous clinical and methodological features are needed to confirm the effectiveness of these interventions on increasing physical activity levels. Validating the evidence for the effects of home or community-based interventions for youth with ID would help clinicians and policymakers create effective frugal programs that increase PA levels in this population and, thus, reduce risks for secondary diseases and improve overall function and quality of life.

Acknowledgments

We would like to acknowledge the assistance of Dr. Amabile Borges Dario in the statistical analyses for this review. This research did not receive any grant from funding agencies.

References

- Krahn GL, Hammond L, Turner A. (2006) A cascade of disparities: health and health care access for people with intellectual disabilities. Mental Retardation and Developmental Disabilities Research Reviews 12(1):70-82.
- Cooper RA, Quatrano LA, Axelson PW, et al. (1999) Research on physical activity and health among people with disabilities: A consensus statement. Journal of Rehabilitation Research and Development 36(2):142-154.
- Carlon SL, Taylor NF, Dodd KJ, et al. (2013) Differences in habitual physical activity levels of young people with cerebral palsy and their typically developing peers: A systematic review. Disability and Rehabilitation 35(8):647-655.
- Lin JD, Lin PY, Lin LP, et al. (2010) Physical activity and its determinants among adolescents with intellectual disabilities. Research in Developmental D is a b i l i t i e s 3 1 (1): 2 6 3 - 2 6 9. https://doi.org/10.1016/j.ridd.2009.09.015
- Emerson E. (2005) Underweight, obesity and exercise among adults with intellectual disabilities in supported accommodation in Northern England. Journal of Intellectual Disability Research 49(Pt 2): 134-143. https://doi.org/10.1111/j.1365-2788.2004.00617.x
- Johnson CC. (2009) The benefits of physical activity for youth with developmental disabilities: A systematic review. American Journal of Health Promotion 23(3):157-167.
- Ouellette-Kuntz H, Garcin N, Lewis MS, et al. (2005) Addressing health disparities through promoting equity for individuals with intellectual disability. Canadian Journal of Public Health/Revue Canadienne de Sante'e Publique. 96(Suppl 2):S8-S22.
- 8. Clemente I. (2017) Barriers and facilitators to participation in physical activity for children with

disabilities: A systematic literature review. Jonkoping University School of Education and Communication. h t t p s : / / w w w . d i v a portal.org/smash/get/diva2:1107967/FULLTEXT01.pdf

- Foley JT, McCubbin JA. (2009) An exploratory study of after-school sedentary behaviour in elementary school-age children with intellectual disability. Journal of Intellectual and Developmental Disability 34(1):3-9.
- Kapsal NJ, Dicke TD, Morin AJS, et al. (2019) Effects of physical activity on the physical and psychosocial health of youth with intellectual disabilities: A systematic review and meta-analyses. Journal of Physical Activity and Health 16(12): 1187-1195. doi: 10.1123/jpah.2018-0675
- 11. Shields N, Taylor NF, Wee E, et al. (2013) A community-based strength training programme increases muscle strength and physical activity in young people with Down syndrome: A randomised controlled trial. Research in Developmental Disabilities 34(12):4385-4394.
- 12. Sevick MA, Dunn AL, Morrow MS, *et al.* (2000) Costeffectiveness of lifestyle and structured exercise interventions in sedentary adults: Results of project ACTIVE. American Journal of Preventive Medicine 19(1):1-8.
- 13. McGarty AM, Downs SJ, Melville CA, *et al.* (2018) A systematic review and meta-analysis of interventions to increase physical activity in children and adolescents with intellectual disabilities. Journal of Intellectual Disability Research 62(4):312-329.
- 14. Liberati A, Altman DG, Tetzlaff J, *et al.* (2009) The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate health care interventions: explanation and elaboration. PLoS Medicine 6(7):e1000100.
- Downs SH, Black N. (1998) The feasibility of creating a checklist for the assessment of the methodological quality both of randomised and non-randomised studies of health care interventions. Journal of Epidemiology & Community Health 52(6):377-384.
- 16. Cohen J. (1988) Statistical power analysis for the behavioral sciences 2nd ed. Hillsdale, NJ: Lawrence Erlbaum Associates.
- 17. Higgins JP, Thompson SG, Deeks JJ, et al. (2003) Measuring inconsistency in meta-analyses. BMJ
 3 2 7 (7 4 1 4) : 5 5 7 - 5 6 0 . https://doi.org/10.1136/bmj.327.7414.557
- 18. Bania TA, Dodd KJ, Baker RJ, *et al.* (2016) The effects of progressive resistance training on daily physical activity in young people with cerebral palsy: A

randomised controlled trial. Disability and Rehabilitation. 38(7):620-626.

- 19. Van Wely L, Balemans AC, Becher JG, *et al.* (2014) Physical activity stimulation program for children with cerebral palsy did not improve physical activity: A randomised trial. Journal of Physiotherapy 60(1):40-49.
- Maher CA, Williams MT, Olds TI, et al. (2010) An internet-based physical activity intervention for adolescents with cerebral palsy: A randomized controlled trial. Developmental Medicine & Child Neurology 52(5):448-455.
- Van den Berg-Emons RJ, Van Baak MA, Speth L, et al. (1998) Physical training of school children with spastic cerebral palsy: effects on daily activity, fat mass and fitness. International Journal of Rehabilitation Research 21(2):179-94.

- 22. Ulrich DA, Burghardt AR, Lloyd M, *et al.* (2011) Physical activity benefits of learning to ride a twowheel bicycle for children with Down syndrome: A randomized trial. Physical Therapy 91(10):1463-1477.
- 23. Hollub AV, Freeman J, Ward S. (2018) The effectiveness of occupational, physical, and speech therapy home health care in early childhood. Pediatrics 141 (1_MeetingAbstract):45. https://doi.org/10.1542/peds.141.1MA1.45
- 24. Shilts MK, Horowitz M, Townsend MS. (2004) Goal setting as a strategy for dietary and physical activity behavior change: a review of the literature. American Journal of Health Promotion 19(2):81-93.

APPENDIX

Appendix A. Sample search strategy using OVID Medline database

Append	ix A. Sample search strategy using OVID Medline database
Databa	se: OvidMEDLINE
1.	Disability/ or Disabled Persons.mp. or Rehabilitation/
2.	exp Developmental Disabilities/
3.	exp Learning Disorders/
4.	exp mental retardation/
5.	exp learning disability/
6.	Intellectual Disability.mp
7.	1 or 2 or 3 or 4 or 5 or 6
8.	(disab* or (special adj1 needs)).ti. or disab*.ab. or (special adj1 needs).ab.
9.	7 or 8
10.	
11.	
	exp Pediatrics/
	10 or 11 or 12 (adolesc* or youth* or child* or teenage* or "young adult*" or paediatric* or pediatric*).ti. or (adolesc* or youth* or child* or
14.	teenage* or "young adult*" or paediatric* or pediatric*).ab.
15.	
-	(home or house or family or families).ab. or home.ti. Or house.ti. or family.ti. or families.ti.
17.	
18.	
-	exp Exercise/
	exp Physical fitness/
	cardiorespiratory fitness.mp.
22.	
23.	exp Child Health Services/ or exp Health Status/ or exp Health Behavior/ or exp Health Promotion/ or exp Adolescent Health/
	or exp Health/
	19 or 20 or 21 or 22 or 23
	Physical activity.mp.
	24 or 25
	exp Clinical Trials/
	exp Randomized Controlled Trials/
	exp Cohort Studies/ exp Case-Control Studies/
	exp Clase-Control Studies/
	exp Pilot Projects/
	exp Single-Blind Method/
	exp Double-Blind Method/
	Cross-Sectional Studies.mp.
	Cross-Over Studies.mp.
	27 or 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36
	(clin* adj25trial*).ti,ab.
	((singl* or doubl* or trebl* or tripl*) adj25 blind*).mp. or mask*.ti,ab.)
	37 or 38 or 39
41.	9 and 15 and 18 and 26 and 40
42.	limit 41 to English
1	

APPENDIX

Appendix B. Methodological quality assessment of included studies

Questions	Bania <i>et al.</i> (2016)	Shields <i>et al.</i> (2013)	Ulrich <i>et al.</i> (2011)	Van Wely <i>et al.</i> (2014)	Maher <i>et al.</i> (2010)	Van Den Berg-Emons e <i>t al.</i> (1998)
1. Is the hypothesis/aim/objective of the study clearly described?	1	1	1	1	1	1
2. Are the main outcomes to be measured clearly described in the Introduction or Methods section?	1	1	1	1	1	1
3. Are the characteristics of the patients included in the study clearly described	1	1	0	1	1	0
4. Are the interventions of interest clearly described?	1	1	1	1	1	1
5. Are the distributions of principal confounders in each group of subjects to be compared clearly described?	2	2	0	2	2	2
6. Are the main findings of the study clearly described?	1	1	1	1	1	1
7. Does the study provide estimates of the random variability in the data for the main outcomes?	1	1	1	1	1	1
8. Have all important adverse events that may be a consequence of the intervention been reported?	1	1	0	1	0	0
9. Have the characteristics of patients lost to follow-up been described?	1	1	1	1	1	1
10. Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?	1	1	1	1	1	0
11. Were the subjects asked to participate in the study representative of the entire population from which they were recruited?	0	0	1	0	1	0
12. Were those subjects who were prepared to participate representative of the entire population from which they were recruited?	0	0	0	1	1	0
13. Were the staff, places, and facilities where the patients were treated, representative of the treatment the majority of patients receive?	1	1	1	0	1	1
14. Was an attempt made to blind study subjects to the intervention they have received?	0	0	0	0	0	0
15. Was an attempt made to blind those measuring the main outcomes of the intervention?	1	1	0	1	1	0
16. If any of the results of the study were based on "data dredging", was this made clear?	1	1	1	1	1	1
17. In trials and cohort studies, do the analyses adjust for different lengths of follow-up of patients, or in case-control studies, is the time period between the intervention and outcome the same for cases and controls?	1	1	1	1	1	1
18. Were the statistical tests used to assess the main outcomes appropriate?	1	1	1	1	1	1
19. Was compliance with the intervention/s reliable?	0	0	1	0	0	1
20. Were the main outcome measures used accurate (valid and reliable)?	1	1	1	1	1	1
21. Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?	0	0	1	0	1	1
22. Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?	0	0	1	0	1	1
23. Were study subjects randomised to intervention groups?	1	1	1	1	1	0
24. Was the randomised intervention assignment concealed from both patients and health care staff until recruitment was complete and irrevocable?	0	0	0	1	1	0
25. Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?	1	1	0	1	1	0
26. Were losses of patients to follow-up taken into account?	1	1	1	1	1	0
27. Did the study explicitly report any methods to obtain power?	1	1	0	1	1	0
Total Quality Score: n% (Highest Possible Score: 28)	21(75)	21(75)	18(64)	22(78)	25(89)	16(57)
Quality Score on Reporting: n% (1-10) (Highest Possible Score: 11)	11(100)	11(100)	7(63)	11(100)	10(90)	8(72)
Quality Score on External Validity: n% (11-13) (Highest Possible Score: 3)	1(33)	1(33)	2(66)	1(33)	3(100)	1(33)
Quality Score on Internal Validity - Bias: n% (14-20) (Highest Possible Score: 7)	5(71)	5(71)	5(71)	5(71)	5(71)	5(71)
Quality Score on Internal Validity - Confounding: n% (21-26) (Highest Possible Score: 6)	3(50)	3(50)	4(66)	4(66)	6(100)	2(33)