THE EFFECTS OF PHYSICAL ACTIVITY AND PHYSICAL EDUCATION ON CHILDREN WITH DOWN SYNDROME: A META-ANALYSIS

By

Christopher Bolt

A Thesis Presented to

The Faculty of Humboldt State University

In Partial Fulfillment of the Requirements for the Degree

Master of Science in Kinesiology: Teaching/Coaching

Committee Membership

Dr. Rock Braithwaite, Committee Chair

Dr. Chris Hopper, Committee Member

Dr. Sean Healy, Committee Member

Dr. Justus Ortega, Graduate Coordinator

July 2017

ABSTRACT

THE EFFECTS OF PHYSICAL ACTIVITY AND PHYSICAL EDUCATION ON CHILDREN WITH DOWN SYNDROME: A META-ANALYSIS

Christopher Bolt

Objective: The purpose of this study was to synthesize findings from physical activity interventions on children and adolescents with Down syndrome.

Design: The present study employed a quantitative research synthesis design. The overall conclusions of past research highlight important issues related to physical activity interventions performed on children and adolescents with Down syndrome.

Methods: Standard meta-analytic procedures incorporating inclusion and exclusion criteria, literature search, coding procedures, and statistical methods were used to identify and synthesize 24 studies with 258 independent samples. Cohen's (1988) criteria for effect sizes were used to interpret and evaluate results.

Results: The average treatment effect for all TARGET intervention studies was moderate ($g = -0.33$; $SE = 0.11$; 95% C.I. = -0.55, -0.11; $p = 0.003$) and represented about 3 tenths of a standard deviation advantage for control groups over the treatment groups. Review of the homogeneity statistics revealed a significant heterogeneous distribution (Q_T =

74.75, $p < 0.05$) making it necessary to explain between-study variation through moderator analyses of characteristics coded for studies. In addition, an outlier analysis was conducted through evaluation of residual values and found one independent sample (Ordonez, 2006) to be an outlier ($z = -5.13$). This prompted the use of a "one-study" removed procedure. The single effect size was retained in the analysis as the results indicated a small change (-0.26) remaining within the 95% confidence interval.

All future quantitative interventions should report all data on all outcomes regardless of their significance level. The most important considerations for the construct of physical activity interventions on children and adolescents with Down syndrome should include the delivery of content from a trained adapted physical education teacher, tasks appropriate for people with Down syndrome, and consistent testing duration to reduce the possibility of physical and cognitive regression. The overall meta-analytic findings indicate that comparing children and adolescents with Down syndrome with their typical developing peers limits our ability to draw firm conclusions on the positive effects of physical activity interventions. More data are needed from the studies to provide a better overall understanding of the current trends in research and application.

TABLE OF CONTENTS

LIST OF TABLES

LIST OF FIGURES

INTRODUCTION

Down Syndrome

The National Birth Defects Prevention Network collects state-specific birth defects data for annual publication of prevalence estimates. Recent studies (Parker et al, 2010) estimate 1 in every 700 child is born with Down syndrome in the United States. Other studies (Maulik et al, 2013) estimate that Down syndrome is one of the most prevalent disabilities in the world affecting 1 in every 964 children who are born. Children with Down syndrome have multiple malformations, medical conditions, and cognitive impairment because of the presence of extragenetic material. (Shieve et al, 2009) The extra genetic material is caused by trisomy of human chromosome 21 (Wiseman et al, 2009). Trisomy is a condition in which an extra copy of a chromosome is present inside of the cell, causing developmental abnormalities.

Down Syndrome and Physical Activity

Physical activity refers to any bodily movement produced by skeletal muscles that results in energy expenditure (Caspersen et al, 1985). Physical activity is necessary in order to prevent many adverse health conditions, including the world's major non-communicable diseases (Lee et al, 2012). The difference in the amount of physical activity that is performed varies from person to person. According to the U.S. Department of Health and Human Services (USDHHS, 2008), it is recommended that

children and adolescents participate in at least 60 minutes of physical activity every day in order to achieve important health benefits (USDHHS, 2008). A recent study found that 58 percent of children with Down syndrome do not meet the daily recommendation of at least 60 minutes of physical activity (Shields et al, 2009).

Down Syndrome and Health Issues

There are specific considerations for people with Down syndrome participating in physical activity. It is important for an individual with Down syndrome to find out if they have atlantoaxial instability before participation in physical activity. Activities and contact sports that put stress on the neck such diving in a pool for example are contraindicated for people with Down syndrome. (Pueschel, 1998). The increased laxity between the first and second cervical vertebrae may sub lux and cause spinal cord injury (Ali et al, 2006) making these activities dangerous. Other developmental abnormalities include musculoskeletal, cardiovascular, intellectual, and biological characteristics (Barr and Shields, 2011). Children with Down syndrome are at risk of hearing loss, eye disease, including cataracts and severe refractive errors, congenital heart defects, neurologic dysfunction, hypermobility, hip dislocation, and lower muscular strength. (Bull, 2011).

Down Syndrome and Public Education

People with disabilities have not always have the right to a free and appropriate education. In the early twentieth century people with disabilities were placed in

institutions, residential programs, and special schools (Polloway et al. 1996). It was not until 1975 when the federal courts interpreted the fourteenth amendment of the constitution to provide education for students with disabilities. The federal courts ruled that a student could not be discriminated against on the basis of a disability and that parents had due process of their children under the Education for All Handicapped Children Act. The Education for All Handicapped Children act was amended in 1997 and is now known as the Individuals with Disabilities Education Act. The Individuals with Disabilities Education Act (IDEA) categorizes Down syndrome as an intellectual disability. This requires students with Down syndrome to receive special education services that include physical education because federal law states that every child has a right to a free and appropriate education. IDEA classifies physical education teachers as direct service providers. The state of California has its own set of law, education codes, and regulations that govern education. The California Code of Regulations (CCR) defines adapted physical education is a program for individuals with exceptional needs who require developmental or corrective instruction and who area precluded from participation in the activities of the general physical education program, modified general physical education program, or in a specially design physical education program in a special class (Adapted Physical Education Guidelines for California Schools, 2012). California Code of Regulations, CCR, Title 6, Section 56363 (a) and (b) (5) classifies physical education teachers as designated instruction and service providers (DIS) and related service providers. (Adapted Physical Education Guidelines for California Schools,

2012). This allows students with Down syndrome to receive instruction from physical education teachers that will allow them to develop skills to build and maintain a physically active lifestyle.

The Individuals with Disabilities Education Act (IDEA) was passed in 1990. IDEA required that all people with disabilities have access to Physical Education in a normal school environment. The problem within this legislation is that each State is left to define what Adapted Physical Education means with respect to complying with the legislation. In response to this legislation, the National Consortium for Physical Education and Recreation for Individuals with Disabilities (NCPEID) developed professional standards and a means of evaluating those standards for Adapted Physical Education in order to determine who is qualified to provide physical education services to students with disabilities. A joint venture by the American Association for Physical Activity and Recreation and National Consortium for Physical Education and Recreation for Individuals with Disabilities produced a document that describes the requisite knowledge and skills that a qualified adapted physical educator must possess to teach in public schools (AAPAR and NCPERID, 2007). It is important to recognize that development of appropriate teaching practices is an ongoing and continuous endeavor just as the development of attributes of an adapted physical educator is an ongoing continuous endeavor (Lytle et al. 2010).

Adapted Physical Education (APE) teachers have a responsibility to ensure that all students are participating in content that addresses specific areas of need, as well as

offering opportunities for students to stress their aerobic and anaerobic body systems with appropriate activities. Evaluation and screening of students with disabilities should be required and performed by a certified APE teacher to obtain a measure of their current physical abilities followed by creating specific goals and objectives that will be incorporated in their physical education program and outlined in the individual's IEP. While there are specific recommendations for adapted physical education programs, teachers, and students with Down syndrome there is little evidence concerning moderating effects. Therefore, the purpose of the following study was to determine the effectiveness of moderating variables on adapted physical education and/or physical activity programs on students with Down syndrome.

METHODS

Search Strategy and Inclusion Criteria

The literature was systematically searched through seven electronic databases from January 1970 until January 2016: Medline, Eric, PsychINFO, PsychARTICLES, Child Development and Adolescent Studies, PubMed, and SPORTDiscus. Combinations of the follow three groups of keywords were used for searching: (i) Down syndrome OR trisomy 21 OR developmental delay OR intellectual delay; AND (ii) physical education OR adapted physical education OR physical activity OR adapted physical activity; AND (iii) evidence based practice OR teaching practice OR teaching strategies OR instructional strategies OR interventions. Articles were searched through the electronic databases and screened by two researchers.

The research process followed this procedure: (i) References were screened first by title, then by abstract, and finally by the entire article; (ii) selected references were downloaded from *Medline, Eric, PsychINFO, PsychARTICLES, Child Development and Adolescent Studies, PubMed,* and *SPORTDiscus* databases and uploaded to Thomson Reuters Endnote X7 software; (iii) Duplicate references were removed from the selection; (iv) Using the Endnote software the references will be analyzed and organized into three folders titled included, excluded, and not sure following the specific inclusion and article selection criteria that is explained below. References sorted into the not sure folder were reanalyzed and reclassified; (v) Copies of the articles based on the references

in the included folder will be downloaded from the 7 electronic databases. Figure 1 provides a diagram summarizing the search process.

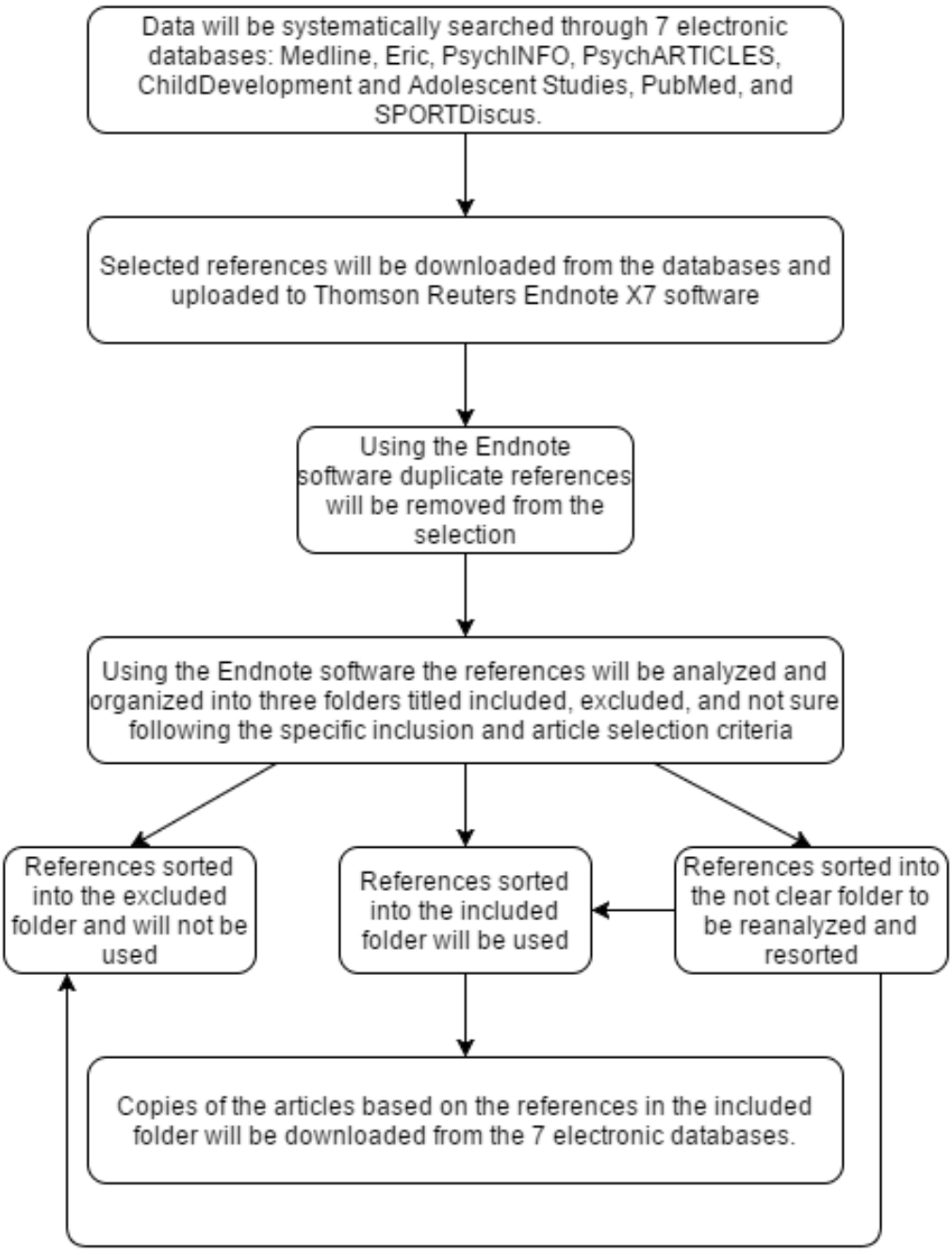

Figure 1. Diagram of Research Process

Reference articles that were included in the review satisfied all of the following criteria: (i) participants are involved in a physical education, physical activity, or a sport setting; (ii) participants are between the ages of 3 and 22; (iii) studies implemented an assessment, method, or intervention in physical education, physical activity, or sport; (iv) studies will have at least one quantitative outcome that has been assessed and reported; (v) studies will be published in the English language; (vi) studies will be published after the year 1970; AND (vii) studies will identify participants with Down syndrome.

Coding Moderating Variables

Information was extracted from each article by two reviewers for demographic information in three subgrouping categories that included *Methodological Characteristics* 1) Design (Descriptive or Experimental); 2) Duration (Unit, Semester, Year); 3) Setting (Inclusive or Specialized Class); 4) Training (Adapted Physical Education, Physical Education, Special Education, Other); 5) Design (Theoretical, A-theoretical); 6) Outcomes (Psychomotor, Cognitive, Affective, Combined). *Sample Characteristics* included 7) Level of Functioning (Mild, Moderate, or Severe); 8) Environment (Physical Activity, Physical Education, or Sport); 9) Gender (Male, Female, Both); 10) School Level (Elementary, Middle, High or Combination); 11) Study Geographical location (Rural or Urban); 12) Country of Origin (US, UK, etc.); and 13) Parent Support (Parental Support OR No Parent Support). *Study Characteristics* included; 14) Study Measure (Objective or Subjective); and 15) Study Status (Published or Unpublished).

Effect Size Calculations

Data were entered into Comprehensive Meta-Analysis version 2 software which was used to compute all effect sizes. Each study was the unit of analysis and contributed one independent effect size to the meta-analysis. The program provided more than 258 data entry options that were used to calculate effect sizes included variations on both matched and unmatched designs across post-test, pre-post contrast and gain scores. Estimates of effects size calculations were based on descriptive statistics such as means, standard deviations, sample sizes, and when necessary *t* or *p* values (Valentine et. al, 2003). Hedges *g* was used as the primary measure of effect, providing a conservative estimate of effect in the smaller sample sizes ($k < 20$) (Hedges & Vevea, 1998). A random effects model was used to model error associated with the current investigation and makes the assumption that there are both within study error and between-study variance that influence the effect size calculation (Bornstein et al., 2009). The rationale for selecting a random effects model was an expected variation between intervention methods, potential sampling error, and the possibility of random unexplained variance between the studies.

Heterogeneity of Variance

When using a random effects model there is an assumption that the true effect size will vary between studies. The *Q*-test serves as a significance test that indicates heterogeneity, or that variability across the effect sizes is greater than what would have resulted from chance. Effect size distributions that are heterogeneous indicate a large variability and allow for study of moderator variables. When interpreting *Q* and

corresponding *p*-values, tau-squared (τ^2) and I-squared (I^2) heterogeneity statistics should be considered for interpretation. The reason for using τ^2 and I^2 is that *p*-values only indicate that true effects vary between studies but do not include information on the magnitude of dispersion. The tau-squared statistic calculates weights and yields an estimate of total variance between studies in a random effects model. Larger tau-squared values indicate the proportion of variance that can be attributed to real differences between the studies. The *I*-squared statistic is the ratio of excess dispersion to total dispersion and can be interpreted as the overlap of confidence intervals. Values closer to zero represent random error and values that move away from zero explain the variance by covariates.

Outlier and Publication Analysis

Outlier analysis were examined by the interpretation of relative residuals and by a "one-study removed" procedure. Any study that was identified as an outlier was examined in a "one-study removed" analysis, studies were not removed if they did not substantially impact the effect size of g and results were within or near the 95^{th} confidence interval. Publication bias was controlled for by a visual inspection of a funnel plot, the Trim and Fill procedure (Duval & Tweedie, 2000) and Fail-Safe-N calculation (Rosenthal, 1979). The funnel plot provided a visual representation of publication bias that was based on a symmetrical distribution of data points about the mean effect size. Symmetrical plots can be interpreted as a lack of publication bias. Asymmetrical data are adjusted by using the Trim and Fill procedure (Duval and Tweedie, 2000). The Trim and

Fill procedure identifies the number of missing studies that would balance the plot to provide an unbiased estimate of effect size. The Fail-Safe-*N* was used to determine the number of non-significant missing studies that would be needed to nullify significant results (Rosenthal, 1979).

RESULTS

The primary purpose of this study was to synthesize findings from physical activity interventions on children and adolescents with Down syndrome in order to determine the overall effectiveness across all outcomes and moderators. The secondary purpose was to determine the effect of physical activity interventions (TARGET) on specific psychomotor outcomes in physical activity settings. The search procedures generated 4469 results. After the screening process, a total of 1026 studies were identified as potential sources for data collection. Data extraction forms following established meta-analytic procedures were then used (Brown et al., 2003) to determine the inclusion status of each study. A total of 24 studies with 24 independent samples were included in the meta-analysis meeting the inclusion criteria. Figure 2 provides an overall presentation of the search strategy in the meta-analysis.

Figure 2. Literature Search Process

Random Effects Model Results

The average treatment effect for all TARGET intervention studies was moderate (g = -0.33; SE = 0.11; 95% C.I. = -0.55, -0.11; p = 0.003) and represented about 3 tenths of a standard deviation advantage for control groups over the treatment groups. Review of the homogeneity statistics revealed a significant heterogeneous distribution (Q_T =

74.75, $p < 0.05$) making it necessary to explain between-study variation through moderator analyses of characteristics coded for studies. In addition an outlier analysis was conducted through evaluation of residual values and found one independent sample (Ordonez, 2006) to be an outlier ($z = -5.13$). This prompted the use of a "one-study" removed procedure. The single effect size was retained in the analysis as the results indicated a small change ($g = -0.26$) remaining significant and within the 95% confidence interval. Publication bias was deemed marginal as a result of a symmetrical funnel plot, no studies being added during the Trim and Fill procedure, and Fail Safe N value calculation of 130 studies that would be needed to nullify a significant α-level ($p < 0.05$).

Moderator Analysis

Heterogeneity statistics for the random effects model confirmed that there was a heterogeneous ($Q_T = 74.75$, $p < 0.05$) distribution and a moderate to large variance ($I^2 = 69.23$) of between study variation existed to justify running a sub-group analyses for coding characteristics. Table 1 presents the results from moderator analyses on study characteristics. All analyses produced overall trends (experimental group < control groups, $p < 0.05$) for specific moderators.

When interpreting the treatment effects Cohen's criteria were used for interpretation of standardized mean differences. Effect sizes of (≤ 0.20) are interpreted as small, (≥ 0.50) as medium, and (≥ 0.80) as large (Cohen, 1988). Positive effect sizes were interpreted as experimental groups having stronger outcome results than control groups. Negative effect sizes are interpreted as control groups having stronger outcome

results than the experimental groups. In all cases moderate to high negative associations were observed with the experimental groups in comparison to the control groups. Overall, the random effects model (g = -0.033, Z = -2.93, p = .003) had a negative moderate effect. The control groups outperformed the experimental groups by more than three tenths of a standard deviation. Table one provides an overview of the moderator results.

Methodological Characteristics

There were not methodological characteristics that were significant between subgroups, however, several trends were apparent in the data. A significant difference was found within the study design moderator. The A-theoretical covariate had a negative moderate effect that was 3 standard deviations away from the mean (g = -0.38, Z = -3.05, p = 0.00). A significant difference was found within the duration moderator and two of the four covariates (less than 3 weeks and months) all had negative moderate effects. Studies conducted in under 3 weeks (g = -0.68, Z = -2.34, p = 0.02) indicate the control groups outperformed the experimental groups by two standard deviations away from the mean. Studies conducted one month to a year (g = -0.63, Z = -2.41, p =0.02) indicate the control groups outperformed the experimental groups by approximately six-tenths of a standard deviation. A significant difference was found within the environment moderator. The physical activity covariate had a negative moderate effect. Studies conducted in a physical activity setting (g= -0.33, Z = -2.83, p = .005) indicate the control groups outperformed the experimental groups by 2 standard deviations away from the mean. A significant difference was found within the outcome moderator. The

psychomotor covariate had a negative moderate effect. Studies that conducted interventions within the psychomotor realm (g= -0.36, Z = -2.89, p = .004) indicate the control groups outperformed the experimental groups by three-tenths of a standard deviation. A significant difference was found within the setting moderator. The "inclusive" covariate had a negative moderate effect. Studies conducted in an inclusive environment (g= -0.50, Z = -3.33, p = .001) indicate the control groups outperformed the experimental groups by one half of a standard deviation. A significant difference was found within the level of functioning moderator. The "not reported" covariate had a negative moderate effect. Studies that did not report a level of functioning for the participants (g= -0.38, Z = -2.98, p = .003) indicate the control groups outperformed the experimental groups by about 3 standard deviations away from the mean.

Sample Characteristics

None of the sample characteristics moderator variables produced significant differences between subgroups, however, trends were apparent in the data. A significant difference was found within the country moderator. The "Spain" covariate had a negative large effect. Studies that were conducted in Spain (g= -0.80, Z = -3.33, p = .000) indicate the control groups outperformed the experimental groups by 3 standard deviations away from the mean. A significant difference was found within the gender moderator. The "males and females" covariate had a negative moderate effect. Studies that were conducted with both males and females (g= -0.49, Z = -3.28, p = .001) indicate the control groups outperformed the experimental groups by approximately half a standard deviation. A significant difference was found within the location moderator.

The “urban” covariate had a negative moderate effect. Studies that were conducted in an urban setting ($g = -0.39$, $Z = -2.81$, $p = .005$) indicate the control groups outperformed the experimental groups by about four-tenths of a standard deviation.

STUDY CHARACTERISTICS

Study characteristics did not produce any significant differences between subgroups, however, there were trends in the data. A significant difference was found within the measure moderator. The objective covariate had a negative moderate effect. Studies conducted in an objective manner (g= -0.36, Z = --2.91, p = .004) indicate the control groups outperformed the experimental groups by almost four-tenths of standard deviation. A significant difference was found within the publication status moderator. The "published" covariate had a negative moderate effect. Studies that were published (g= -0.34, Z = -2.86, p = .004) indicate the control groups outperformed the experimental groups by three-tenths of a standard deviation. A significant difference was found within the support moderator. The "not reported" covariate had a negative moderate effect, and the "parental support' covariate had a negative large effect. Studies conducted without reporting parental support (g= -0.95, Z = -2.92, p = .004) indicate the control groups outperformed the experimental groups by approximately one standard deviation. Studies conducted with parental support (g= -0.25, Z = -2.10, p = .04) indicate the control groups outperformed the experimental groups by two-tenths of standard deviation.

Table 1. Subgroup Analyses

	k	*g*	*SE*	s^2	95% C.I.	*Z*	*Q*	τ^2	I^2	Fail Safe N
Random Effects Model [a]	24	-0.33	0.11	0.001	(-0.55, -0.11)	-2.93*	74.75*	0.19	69.23	130
Methodological Characteristics [b]										
Design							1.14			
A theoretical	22	-0.38	0.12	0.02	(-0.62, -0.14)	-3.05*	71.45*	0.22	70.61	
Theoretical	2	0.04	0.37	0.14	(-0.69, 0.76)	0.11	0.30	0.00	0.00	
Duration							5.31			
< 3 Weeks	4	-0.68	0.29	0.08	(-1.24, -0.11)	-2.34*	8.88*	0.29	66.20	
Months	5	-0.63	0.26	0.07	(-1.14, -0.12)	-2.41*	27.20*	0.74	85.29	
Not Reported	13	-0.24	0.15	0.02	(-0.53, 0.06)	-1.58	23.90*	0.08	49.79	
Years	2	0.17	0.34	0.11	(-0.49, 0.82)	0.49	1.02	0.00	1.62	
Environment							0.04			
Physical Activity	23	-0.33	0.12	0.01	(-0.56, -0.10)	-2.83*	74.50*	0.19	70.47	
Physical Education	1	-0.46	0.64	0.41	(-1.71, 0.80)	-0.72	0.00	0.00	0.00	
Level of Functioning							0.76			
Mild to Moderate	3	-0.08	0.31	0.10	(-0.70, 0.53)	-0.27	0.16	0.00	0.00	
Not-Reported	21	-0.38	0.13	0.01	(-0.63, -0.13	-2.98*	72.34*	0.25	72.35	
Outcome							0.55			
Cognitive	1	-0.46	0.65	0.42	(-1.73, 0.081)	-0.71	0.00	0.00	0.00	
Combination	2	-0.05	0.41	0.17	(-0.86, 0.77)	-0.11	0.69	0.00	0.00	
Psychomotor	21	-0.36	0.12	0.02	(-0.60, -0.12)	-2.89*	73.39*	0.21	72.75	
Setting							2.78			
Inclusive	12	-0.50	0.15	0.02	(-0.80, -0.21)	-3.33*	15.55	0.05	29.28	
Specialized	12	-0.16	0.14	0.02	(-0.44, 0.12)	-1.14	41.73*	0.19	73.64	
Sample Characteristics [b]										
Age level							1.26			
Combined	9	-0.43	0.18	0.03	(-0.78, -0.09)	-2.44*	33.76*	0.18	76.30	
Elementary School	8	-0.14	0.21	0.05	(-0.56, 0.28)	-0.64	14.04*	0.14	50.15	
High School	7	-0.41	0.23	0.06	(-0.87, 0.05)	-1.75	24.56*	0.37	75.60	
Country							9.57			
Brazil	1	.045	0.48	0.24	(-0.50, 1.40)	0.93	0.00	0.00	0.00	
Egypt	1	-0.13	0.50	0.25	(-1.12, 0.85)	-0.26	0.00	0.00	0.00	

	k	*g*	*SE*	s^2	95% C.I.	*Z*	*Q*	τ^2	I^2	Fail Safe N
Random Effects Model [a]	24	-0.33	0.11	0.001	(-0.55, -0.11)	-2.93*	74.75*	0.19	69.23	130
Europe	1	0.08	0.79	0.62	(-1.47, 1.62)	0.10	0.00	0.00	0.00	
Iran	1	-1.09	0.61	0.37	(-2.27, 0.10)	-1.80	0.00	0.00	0.00	
Japan	1	-0.50	0.64	0.41	(-1.76, 0.76)	-0.78	0.00	0.00	0.00	
Poland	1	-0.46	0.64	0.40	(-1.70, 0.79)	-0.72	0.00	0.00	0.00	
Romania	1	0.01	0.48	0.23	(-0.93, 0.96)	0.02	0.00	0.00	0.00	
Spain	5	-0.80	0.24	0.06	(-1.28, -0.33)	-3.33*	26.76*	0.42	85.05	
Gender							4.26			
Males	8	-0.31	0.19	0.04	(-0.69, 0.60)	-1.64	26.74*	0.27	73.82	
Males and Females	11	-0.49	0.15	0.02	(-0.79, -0.20)	-3.28*	18.39*	0.07	45.51	
Not Reported	5	0.08	0.24	0.06	(-0.38, 0.54)	0.35	10.82*	0.18	63.04	
Location							1.56			
Not Reported	6	-0.10	0.24	0.06	(-0.56, 0.36)	-0.42	1.70	0.00	0.00	
Rural	1	-0.63	0.48	0.23	(-1.58, 0.31)	-1.32	0.00	0.00	0.00	
Urban	17	-0.39	0.14	0.02	(-0.67, -0.12)	-2.81*	65.78	0.28	75.66	
Study Characteristics [b]										
Measure							0.30			
Combined	3	-0.16	0.34	0.11	(-0.82, 0.50)	-0.47	1.23	0.00	0.00	
Objective	21	-0.36	0.12	0.02	(-0.60, -0.12)	-2.91*	73.39*	0.21	72.25	
Publication Status							0.04			
Published	23	-0.34	0.12	0.01	(-0.57, -0.11)	-2.86*	74.75*	0.20	70.57	
Unpublished	1	-0.23	0.54	0.29	(-1.28, 0.81)	-0.44	0.00	0.00	0.00	
Support							4.09*			
Not Reported	4	-0.95	0.32	0.11	(-1.58, -0.31)	-2.92*	19.96*	1.37	84.97	
Parental	20	-0.25	.012	0.01	(-0.48, -0.02)	-2.10*	48.45*	0.11	60.79	

Note. *k* = number of effect sizes. *g* = effect size (Hedges g). *SE* = standard error. S2 = variance. 95% *C. I.* = confidence intervals (lower limit, upper limit). *Z* = test of null hypothesis. τ^2 = between study variance in random effects model. I^2= total variance explained by moderator. * indicates $p < .05$. a = Total *Q*-value used to determine heterogeneity. b = Between *Q*-value used to determine significance ($\alpha < 0.05$)

DISCUSSION

The purpose of this study focused on the effectiveness of physical activity interventions on children and adolescents with Down syndrome and moderating factors that contributed to positive or negative results. In this systematic review and meta-analysis, we observed a significant, moderate to large, negative treatment effect for participants in the experimental groups. We observed a non-significant, small to moderate, positive treatment effect for participants in the control groups. Children and adolescents with Down syndrome who participated in physical activity interventions did not perform better than their counterparts.

Currently, there is a lack of research explicitly focused on physical activity and physical education and Down syndrome that provides a framework to support evidence based practice. This was evident in the number of studies using theoretical design of the articles we examined as 91 percent of the articles included in our meta-analysis did not use a theoretical design. More research needs to be conducted to formulate theories to explain, predict, and understand the phenomena being observed.

The duration between each of the tests that occurred may also significantly affect the results for people with Down syndrome. When comparing people with Down syndrome to their typically developing peers it is shown they performed worse over time. Carlesimo and colleagues (1997) provide a possible explanation due to children with Down syndrome's inability to use explicit memory which helps their intentional recollection previous experiences and concepts (Ullman, 2004). The lack of a positive

effect prevents any conclusions from being drawn on a duration that would facilitate positive outcomes for individuals with Down syndrome. More specifically, 54 percent of studies did not report a study duration that would provide insight into time periods that would facilitate positive effects and when combined with comparisons of groups with and without Down syndrome recommendations are not possible. Providing time references applies to physical education because people with Down syndrome have memory complications (Vicari et al, 2000). If information on the amount of time in between activities is not documented teachers will not be able to design appropriate instruction to facilitate individuals with Down syndrome memory and recall capabilities that will enhance knowledge and skills for lifetime activity.

Ninety six percent of the research was performed in a physical activity setting such as a lab or clinic as compared to four percent of the data being record was performed in practical settings such as a physical education classroom. A setting that is unfamiliar such as a lab or clinic could negatively affect the gross and fine motor function (Blain et al, 1998). A setting that is familiar and has a daily routine such as a physical education classroom may be a better testing environment to get results that are more accurate. More information on practical settings will provide a foundation on how dynamic settings influence outcomes in individuals with Down syndrome. Furthermore, a setting that is familiar relates to physical education and is similar to several activity settings when compared to labs or clinics that might be ideal for testing but not an stable and familiar environment. The type of environment is increasingly important because it

has been shown as people with Down syndrome age, they get even less exercise (Graham & Reid, 2000) which can impair long-term self-care activities.

Eighty six percent the data intake was objective and only three articles had combined both subjective and objective data, and 85 percent of the studies were conducted on psychomotor outcomes. Most of the information from objective measures was based on psychomotor outcomes and combined measures using subjective data came from the parents of the children and adolescents with Down syndrome. These findings are problematic for several reasons. First, self-perceived questionnaires do not accurately represent health and physical activity levels as a study by Hedov, Annerén, and Wikblad (2000) revealed mothers inaccurately described health and physical activities. Triangulating parental physical activity information with students self-perceived questionnaires and corresponding fitness levels provides objective results to design appropriate levels of activity. Second, given the longer duration in between studies could have had a significant negative effect on the psychomotor results because it directly relates to the origination of movement in conscious mental activity (Ullman, 2004) that can decline over time. Developmental aspects such as brain characteristics, musculoskeletal abnormalities, associated medical conditions, inadequate physical activity levels (recommended 60 minutes of moderate to vigorous physical activity) (Esposito al, 2012) are the most significant factors that affect psychomotor development (Martinez et al, 2008) and most likely account for the difference in results when comparing children with Down syndrome and their typical developing peers.

Eighty three percent of the studies that were conducted involved parental support, however, none of the studies mentioned whether the parents had any formal training related to physical education/activity and Down syndrome. Parents are key support agents in their children's life and it has been proven that parental intervention in physical activity and physical therapy can lead to earlier motor development and improved fitness levels (Ulrich et al, 2001). However, providing services that requires specialized training or expertise should be conducted by professionals who are qualified such as a certificated adapted physical education teacher (NASPE, 2007). Combining highly qualified teachers with parents can promote positive educational outcomes. Individualized education plans involve a team of professionals including parents and the physical education teacher (Rupar et al, 2011). Parents are familiar with their children's pediatricians, doctors, and overall health history (Bull, 2011) so it is important to interact with them frequently to stay continually updated on current health information for each student.

The two settings evaluated in the studies were inclusive environments and specialized environments. Fifty percent of the studies were conducted in an inclusive environment where the Down syndrome subjects performed the same tests alongside of their typically developing peers. The other half of the studies were conducted in a specialized environment where subjects with Down's syndrome were tested away from their typically developing peers. In both environments subjects with Down syndrome performed worse than their typically developing peers but the inclusive environment had a significant negative effect. Unfortunately, these comparisons do not allow for any

conclusions concerning the effects on outcomes during inclusive or specialized environments. There are positives to inclusive environments such as peer tutors and the development of friendships between students with and without disabilities (Houston-Wilson et al, 1997) but students with Down syndrome may perform worse in this environment if there aren't teacher assistants, specialists, aids, and peer tutors that are properly trained. It is shown that with proper support systems the effects in physical education are positive (Long et al, 1980).

The aim of our meta-analysis was to collect data from published and unpublished literature but most data conducted on individuals with intellectual disabilities occurs in developed countries (Maulik et al, 2011) making it hard to get an accurate representation of the global population. While the United States of America had the most research to draw conclusions from, Spain was second and had the most significant negative effect. More information is needed that would allow for a better understanding on how cultural perspectives facilitate improvements in both structured and unstructured physical activity.

Most of studies conducted did not determine or report a level of functioning, female-only samples, and information on age-related outcomes. This makes it impossible to draw conclusions from the information for specific levels of functioning related to both skill and age as well as gender. Level of functioning is important because it is an indication of previous medical management, home environment, early intervention, education, and vocational training that each subject may have experienced (Bull, 2011). This relates to physical education because working memory and long-term memory are important considerations (Pennington et al, 2003) when developing activities that

promote fitness, but not every person functions at the same level. Therefore, information is needed so best practices and teaching methods can be developed for each specific level.

CONCLUSION

When analyzing the physical activity interventions, more data are needed from the studies to provide a better overall understanding of the current trends in research and application. All future quantitative interventions should report all data on all outcomes regardless of their significance level. The most important considerations for the construct of physical activity interventions on children and adolescents with Down syndrome should include the delivery of content from a trained adapted physical education teacher, tasks appropriate for people with Down syndrome, and consistent testing durations to reduce the possibility of physical and cognitive regression.

The overall meta-analytic findings indicate that comparing children and adolescents with Down syndrome with their typical developing peers limits our ability to draw firm conclusions on the positive effects of physical activity interventions. Interventions should be conducted using specific strategies that address the unique needs for people with Down syndrome. It is the responsibility of researchers and professionals to provide empirical evidence that demonstrates the effectiveness of specific learning environments and instructional approaches so teachers and other researchers can utilize best practices (Valentini & Rudisill, 2004; Buckley et al, 2006). The findings of this meta-analysis show that more research needs to be conducted in inclusive and specialized physical education environments. Level of functioning is a determining factor for specialized and inclusive environments (Pivik et al, 2002). Therefore, it is necessary to report level of functioning and learning environment in all future studies. The data can

be compared and inferences can be made on the best learning environments for each level of functioning. Additionally, age as it relates to school level can be compared with that data to make long term decisions on learning environments and teaching practices for in education.

RECOMMENDATIONS FOR FUTURE RESEARCH

Several methodological features were explored in an attempt to explain current findings. With regard to the duration of the studies we observed, many of them spanned different time frames and over half did not report a time frame. Children and adolescents with Down syndrome have cognitive development and memory complications (Einfield et al., 2006; Rimmer et al., 2011); Martin et al., 2009) that could be a factor for consistent testing over longer durations of time. More information and consistent testing durations are needed in future research in order to better assess the effect of physical activity interventions on children and adolescents with Down syndrome. With regard to environment and outcome, all but one of the studies were performed in a physical activity setting, and all but 3 interventions were focused on the psychomotor realm. Physical activities interventions should be delivered by certified adapted physical education teachers (Shields et al., 2009; Lee et al., 2012) and the activities being performed should be recommended (Biddle et al., 2000; Minor & Brown, 1993; Rall & Roubenoff, 1996; French et al., 1992; Hunt, 2003; Merriman, 1996; King & Mace, 1990) for children and adolescents with Down syndrome. Any support that is given should be documented and included in the study to determine if it had an overall effect on the results. The link between inclusive and specialized settings in the environment should be noted as well. In inclusive environments participants with Down syndrome are working in conjunction with typically developing participants. The delivery of content and intervention selection (Roizen, 2011; Bull, 2011; Karmiloff-Smith, 2016) should address each participant's

individual developmental level and needs. Even though it will take considerable time and resources to locate and obtain data from unpublished sources it could make a significant difference in our understanding of best practices for people with Down syndrome as it relates to physical activity and exercise.

Most of the studies came from United States of America and Spain and were performed in an urban location. A more diverse population of studies will give more data to analyze current trends in Down syndrome on a global scale. Only three studies reported the current level of functioning of the participants. When focusing on a target population that is diverse, it will be important to report the level of functioning of the participants so patterns and trends can be recognized to improve and develop best teaching practices. (Lytle et al. 2010; IDEA, 2007; Martin et al. 1996; Turnbull et al. 2004). Additionally, age appropriate tasks and interventions should be implemented. Participants with Down syndrome performed the worse in studies that had combined age levels. This could be a result of developmental delays and age appropriate psychomotor interventions (Mehdian, & Kerslake, 2008; Rimmer et al. 2004, Dobbins et al. 1981; Fernhall and Tymeson, 1988; Suomi and Koceja, 1994).

REFERENCES

Adapted Physical Education Guidelines for California Schools, 2012. California Department of Education, Special Education Division

Ali FE, Al-Bustan MA, Al-Busairi WA, Al-Mulla FA, Esbaita EY. Cervical spine abnormalities associated with Down syndrome. Int Orthop (SICOT) 2006;30:284–289.

American Association for Physical Activity and Recreation & National consortium for Physical Education and Recreation for Individuals with Disabilities. (2007). *Highly qualified adapted physical education teacher.*

Biddle, S.J.H., Fox, K.R., and Boutcher, S.H. (2000) Physical Activity and Psychological Well-Being. Routledge, London.

Blain, Tracey and Angel, Jean, "The Effect of Familiar and Novel Testing Environments on the Gross Motor Function Measure Scores of Developmentally Normal Children 17-37 Months Old" (1998).Masters Theses. 395. http://scholarworks.gvsu.edu/theses/395

Buckley, S., Bird, G., & Sacks, B. (2006). Evidence that we can change the profile from a study of inclusive education. *Down syndrome Research and Practice, 9*(3), 51-53.

Bull, Marilyn J. "Health supervision for children with Down syndrome." *Pediatrics* 128.2 (2011): 393-406.

Burdett, S., Stewart, L., & Tierney, J. (2003). PUBLICATION BIAS AND META-ANALYSES: A Practical Example. *International Journal of Technology Assessment in Health Care, 19*(1), 129-134. doi:10.1017/S0266462303000126

Carlesimo, G. A., Marotta, L., & Vicari, S. (1997). Long-term memory in mental retardation: evidence for a specific impairment in subjects with Down's syndrome. *Neuropsychologia*, *35*(1), 71-79.

Caspersen CJ, Powell KE, Christenson GM. Physical activity, exercise, and physical fitness: definitions and distinctions for health-related research. Public Health Rep. 1985;100(2):126–131.

Dobbins, A.D., Garron, R., and Rarick, G.L. (1981). The motor performance of educable mentally retarded and intellectually normal boys after covariate control for differences in body size. Res. Q. Exerc. Sport 52, 1–8.

Duval, S. J., & Tweedie, R. L. (2000). Trim and fill: A simple funnelplot-based method of testing and adjusting for publication bias in metaanalysis. Biometrics, 56(2), 455– 463.

Easterbrook, P. J., Gopalan, R., Berlin, J. A., & Matthews, D. R. (1991). Publication bias in clinical research. *The Lancet*, *337*(8746), 867-872.

Esposito, P. E., MacDonald, M., Hornyak, J. E., & Ulrich, D. A. (2012). Physical activity patterns of youth with Down syndrome. *Intellectual and developmental disabilities*, *50*(2), 109-119.

Evans, D. W., & Gray, F. L. (2000). Compulsive- like behavior in individuals with Down syndrome: its relation to mental age level, adaptive and maladaptive behavior. *Child Development*, *71*(2), 288-300.

Fernhall, B. and Tymeson, G.T. (1988).Validation of a cardiovascular fitness field test for adults with mental retardation. Adapted Phys. Act. Q. 5, 49–59.

French, R., Silliman, L.M., Ben-Ezra, V., and Landrien-Seiter, M. (1992) Influence of selected reinforcers on the cardiorespiratory exercise behavior of profoundly mentally retarded youth. Percept. Mot. Skills 74, 584–586

Hedov, G., Annerén, G., & Wikblad, K. (2000). Self-perceived health in Swedish parents of children with Down's syndrome. *Quality of life research*, *9*(4), 415-422.

Houston-Wilson, C., Dunn, J. M., Mars, H. V. D., & McCubbin, J. (1997). The effect of peer tutors on motor performance in integrated physical education classes. *Adapted Physical Activity Quarterly*, *14*(4), 298-313.

Hunt, A. (2003) Musculoskeletal fitness: the keystone in overall well-being and injury prevention. Clin. Orthop. (409), 96–105.

Individuals with Disability Education Act Amendments of 1997 [IDEA].

Martin, E., Martin, R., & Terman, D. (1996). The legislative and litigation history of special education. *The Future of Children, 6*, 25-39.

Karmiloff-Smith A, Al-Janabi T, D'Souza H *et al.* The importance of understanding individual differences in Down syndrome version 1; 2016

King, D. and Mace, E. (1990). Acquisition and maintenance of exercise skills under normalized conditions by adults with moderate and severe mental retardation. Ment. Retard. 28(5), 311–317.

Lee, I.-M., Shiroma, E. J., Lobelo, F., Puska, P., Blair, S. N., & Katzmarzyk, P. T. (2012). Impact of Physical Inactivity on the World's Major Non-Communicable Diseases. *Lancet*, *380*(9838), 219–229. http://doi.org/10.1016/S0140-6736(12)61031-9

Long, E., Irmer, L., Burkett, L.N., Glasenapp, G., & Odenkirk, B. (1980) PEOPLE. *Journal of Physical Education and Recreation*, 28-29.

Lytle, R, Lavay, B, Rizzo, T. What is a highly qualified adapted physical education teacher? Journal of Physical Education, Recreation and Dance. 2010; 81(2):40-50.

Martin, G. E., Klusek, J., Estigarribia, B., & Roberts, J. E. (2009). Language characteristics of individuals with Down syndrome. *Topics in Language Disorders*, *29*(2), 112–132.

Martinez, N. B., & García, M. M. (2008). Psychomotor development in children with Down syndrome and physiotherapy in early intervention. *International Medical Review on Down Syndrome*, *12*(2), 28-32.

Maulik, P. K., Mascarenhas, M. N., Mathers, C. D., Dua, T., & Saxena, S. (2011). Prevalence of intellectual disability: a meta-analysis of population-based studies. *Research in developmental disabilities*, *32*(2), 419-436.

Merriman, W.J., Barnett, B.E., and Jarry, E.S. (1996). Improving fitness of dually diagnosed adults. Percept. Mot. Skills 83(3 Pt 1), 999–1004.

Minor, M.A. and Brown, J.D. (1993) Exercise maintenance of persons with arthritis after participation in a class experience. Health Educ. Q. 20, 83–95.

Parker SE, Mai CT, Canfield MA, Rickard R, Wang Y, Meyer RE, et al. Updated national birth prevalence estimates for selected birth defects in the United States, 2004–2006. 2010;88:1008–1016.3

Pennington, B. F., Moon, J., Edgin, J., Stedron, J., and Nadel, L. (2003). The neuropsychology of Down syndrome: evidence for hippocampal dysfunction. Child Develop. 74:75–93.

Pivik, J., McComas, J., & Laflamme, M. (2002). Barriers and facilitators to inclusive education. *Exceptional children*, *69*(1), 97-107.

Rimmer JH, Riley B, Wang E, Rauworth A, Jurkowski J. Physical activity participation among persons with disabilities: barriers and facilitators. *Am J Prev Med* 2004; 26: 419–25.

Ruppar, Andrea L., and Janet S. Gaffney. "Individualized education program team decisions: A preliminary study of conversations, negotiations, and power." *Research and Practice for Persons with Severe Disabilities* 36.1-2 (2011): 11-22.

Shields N, Dodd KJ, Abblitt C. Do children with down syndrome perform sufficient physical activity to maintain good health? A pilot study. Adapted Physical Activity Quarterly. 2009;26(4):307–20.

Suomi, R. and Koceja, D.M. (1994) Postural sway patterns of normal men and women and men with mental retardation during a two-legged stance test. Arch. Phys. Med. Rehabil. 75(2), 205–209.

Turnbull, H., Huerta, N., & Stowe, M. (2004). *The Individuals with Disabilities Education Act as Amended in 2004*. Upper Saddle River, New Jersey: Pearson Education, Inc.

Ullman, MT (2004). "Contributions of memory circuits to language: the declarative/procedural model". *Cognition*. 92: 231–70. doi:10.1016/j.cognition.2003.10.008

Ulrich, D. A., Ulrich, B. D., Angulo-Kinzler, R. M., & Yun, J. (2001). Treadmill training of infants with Down syndrome: evidence-based developmental outcomes. *Pediatrics*, *108*(5), e84-e84.

U.S. Department of Health and Human Services. *2008 Physical Activity Guidelines for Americans.* Washington, DC: U.S. Department of Health and Human Services; 2008.

Valentini, N. C., & Rudisill, M. E. (2004). An inclusive mastery climate intervention and the motor skill development of children with and without disabilities. *Adapted physical activity quarterly*, *21*(4), 330-347.

Vicari, S., Bellucci, S., and Carlesimo, G. A. (2000). Implicit and explicit memory: a functional dissociation in persons with Down syndrome. Neuropsychologia 38:240–251.

Wiseman FK, Alford KA, Tybulewicz VL, Fisher EM. Down syndrome–recent progress and future prospects. Human Molecular Genetics. 2009;18(R1):R75–R83.