THE RELATIONSHIP BETWEEN EXERCISE AND EXECUTIVE FUNCTION IN INDIVIDUALS WITH DOWN SYNDROME

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ABSTRACT

Executive function is a set of processes that are responsible for organizing and controlling goal oriented behavior, especially in novel situations (Banich, 2009) and there is mounting evidence that exercise improves executive function. It is also a known deficit in individuals with Down syndrome. The current study investigated if there is a relationship between exercise and executive function in Down syndrome. Participants were children and adults ages 6-18 who have previously been diagnosed with Down syndrome. We found no relationship between exercise and executive function, but many important descriptive findings emerged from the study. Specifically, the current study includes novel information related to average steps per day in a population in Down syndrome, replicated the executive function profile found in previous research, and contributes many important methodological suggestions for future studies investigating the relationship between exercise and executive function in Down syndrome.

DEDICATION

This thesis is dedicated to the families and communities willing to participate in my project. It is also dedicated to all who guided me though the trials and tribulations of creating this manuscript.

LIST OF ABBREVIATIONS AND SYMBOLS

df	Degrees of freedom: number of values free to vary after certain restrictions have been placed on the data
p	Probability associated with the occurrence under the null hypothesis of a value as extreme as or more extreme than the observed value
r	Pearson product-moment correlation
t	Computed value of a *t* test
$<$	Less than
$=$	Equal to
$\geq$	Greater than or equal to

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1. INTRODUCTION

Down Syndrome

According to recent estimates, approximately 1 out of every 700 babies are born with Down syndrome (DS) in the United States (Parker et al., 2010). The trisomy of chromosome 21 is the cause of approximately 95% of diagnoses with DS, translocation of the extra chromosome is the cause of approximately 3% of diagnoses, and mosaic DS is the cause of approximately 2% of the diagnoses (Shin, Siffel, & Correa, 2010). It is important to note that despite the specific cause of DS, the pervading characteristics present in individuals with DS remain the same. Specifically, the overexpression of chromosome 21 causes both physical and mental differences in people with DS.

Physically, those with DS have particular characteristics that make them distinguishable from those without DS. Some common characteristics are a flattened face, almond-shaped eyes that slant up, a short neck, small ears, a tongue that tends to stick out of the mouth, small hands and feet, poor muscle tone, and short stature ("CDC - Birth Defects, Down Syndrome - NCBDDD," n.d.). Individuals with DS also display differences in cognitive development when compared to typically developing individuals (i.e., they have intellectual disability (ID)). Those with DS exhibit relative weaknesses with expressive language, syntactic/morphosyntatic processing, and verbal working memory when compared to others with conditions resulting in intellectual disability (e.g., Abbeduto, Warren, & Conners, 2007; Silverman, 2007). The proposed study does not aim to measure broad cognition as much as it aims to measure executive function—an important subcategory of cognitive processes.

Executive Function

Executive function is a term that encompasses several related, but distinct cognitive processes. Together these processes are responsible for organizing and controlling goal oriented behavior, especially in novel situations (Banich, 2009; Lezak, 1982; Zelazo, Carter, Reznick, & Frye, 1997). Three of the more basic processes of executive function are inhibition, set shifting, and working memory (Miyake et al., 2000; Zelazo, Craik, & Booth, 2004). Inhibition refers to a person's ability to stop, or inhibit, a behavior or a thought. Set shifting, also called attention shifting or cognitive flexibility, refers to a person's ability to switch their attention from one task to another. Working memory refers to a person's capacity to hold information in their mind and simultaneously manipulate it. It is common for these different processes to be studied individually when evaluating executive function (EF) because there is no single test that accurately measures all the processes involved.

Executive Function in Down Syndrome. Executive function deficit is a known characteristic of DS (Carney, Brown, & Henry, 2013; Lanfranchi, Jerman, Dal Pont, Alberti, & Vianello, 2010; Lee et al., 2011; Rowe, Lavender, & Turk, 2006). For example, it has been found that there is a general deficit in inhibitory control in those with DS (Borella, Carretti, & Lanfranchi, 2013). In a study involving both participants with DS and typically developing (TD) participants matched for mental age, participants underwent a battery of EF tasks (Lanfranchi, Jerman, Dal Pont, Alberti, & Vianello, 2010). The researchers found that participants with DS performed significantly more poorly than TD participants on tasks assessing set shifting, planning/problem solving, working memory, and inhibition/preservation, but performed similarly on tasks assessing fluency. Others have also identified working memory as a specific deficit in DS–especially verbal working memory (Baddeley & Jarrold, 2007; Rowe et al., 2006; Silverman, 2007).

A deficit in EF affects all areas of life. For example, Lezak (1982) noted that impairments in EF compromise a person's ability to maintain an independent, constructively self-serving, and socially productive life. Executive function was found to be an indicator of how well children adjust to middle school (Jacobson, Williford, & Pianta, 2011) and is also associated with the development of social skills (Kiley-Brabeck & Sobin, 2006). Given the documented difficulties in EF in DS and the importance of EF to everyday functioning, it is clear that those with DS would benefit from improving EF.

In TD and ID populations, interventions to improve EF have become of increasing interest, and the success of the interventions has varied. There have been a few interventions that have effectively used cognitive training to improve EF, (e.g. Dahlin, Bäckman, Neely, & Nyberg, 2009; Röthlisberger & Neuenschwander, 2012) especially aspects of working memory. Recent intervention studies have recognized and built on the ability to train the processes associated with EF as a way to improve EF, or maintain it while aging (Dorbath, Hasselhorn, & Titz, 2013; Zinke et al., 2014). A study conducted by Miranda et al. (2011) implemented an intervention for children with ADHD using small-group teaching techniques for parents and children with training tasks related to EF (i.e., self-instruction techniques, problem solving, anger management, etc.). After a 10-week intervention period the investigators concluded that the interventions could have a positive effect on some domains of EF.

Executive Function and Exercise. The relationship between exercise and EF has been the subject of an increasing amount of research in recent years. Many studies have focused on the typically developing population, with the bulk of the research focusing on children and older adults (Boucard et al., 2012; Chang, Liu, Yu, & Lee, 2012; Davis et al., 2011; Tanaka et al., 2009; Tomporowski, Davis, Miller, & Naglieri, 2008; Verbeken, Braet, Goossens, & van der Oord, 2013). A recent review article on exercise and EF highlighted many studies that provide evidence that aerobic exercise promotes children's effortful and goal oriented cognition (see Best, 2010). Interestingly, the reported effects have been detected after both acute bouts of exercise as well as after chronic physical training. For example, Davis et al. (2011) conducted an intervention study of overweight children (BMI ≥ 85%, age 7-11) in which they assessed changes in cognitive function between children who were assigned to a no exercise control, a 20-min aerobic exercise group, or a 40-min aerobic exercise group every school day. Cognitive functioning was assessed pre and post the 3-month intervention. They measured four aspects of cognitive processes, though planning was the only one related to EF. They found that both conditions of aerobic training had a positive effect on tasks requiring planning compared to the no exercise condition. More specifically, the subscale of planning included strategy generation and application, self-regulation, intentionality, and utilization knowledge. Acute exercise has been associated with an increased ability to store memories and affect cognitive health in preadolescents (Hillman et al., 2009; Pesce, Crova, Cereatti, Casella, & Bellucci, 2009). Hillman et al. (2009) noted that acute exercise affects underlying processes that support EF across the lifetime.

Neural Mechanisms. There are several possible mechanisms by which exercise may impact EF skills. A number studies have shown that exercise produces enduring positive effects on the brain (Cotman, Berchtold, & Christie, 2007; Dishman et al., 2006; Erickson et al., 2011; Kramer & Erickson, 2007; Praag, Christie, Sejnowski, & Gage, 1999; Praag, 2009). More specifically, exercise increases blood flow to the brain (see Best, 2010; Erickson et al., 2011;

Laurin, Verreault, Lindsay, MacPherson, & Rockwood, 2001), increases levels of brain-derived neurotrophic factor (BDNF) which encourages neurogenesis (Best, 2010; Cotman et al., 2007; Dishman et al., 2006), and may increase cortical plasticity (Cotman et al., 2007; Dishman et al., 2006; Kramer & Erickson, 2007; Praag, 2009). Further, exercise may impact hippocampal size, which correlates with performance on executive function task across species (Schultz & Dunbar, 2010). In one study, Erickson et al. (2011) implemented a 12-month aerobic and stretching exercise intervention in community-dwelling older adults. The aerobic exercise group experienced a 2% increase in hippocampus size whereas the stretching control group experienced a 1.4% decrease in hippocampus size. This is especially relevant considering those with Down syndrome have a proportionally smaller hippocampus (Anderson et al., 2013).

Down Syndrome and Exercise. The relationship between DS and exercise has only recently gained the attention of cognitive researchers. In fact, much of the current literature on exercise in DS does not examine the cognitive benefits of exercise. Rather, the research in this area focuses on how exercise affects bone mass (González-Agüero et al., 2012) and cardiovascular health (Giagkoudaki, Dimitros, Kouidi, & Deligiannis, 2010; Mendonca, Pereira, & Fernhall, 2013) as well as improvements in leg strength and general physical fitness (Cowley et al., 2011; Rimmer, Heller, Wang, & Valerio, 2004). Hinckson and Curtis (2013) recently reviewed the literature on physical activity and its measurement in those with ID. From the 30 articles reviewed, overall, children with ID were significantly less active than those without disabilities. To date, very few studies examine the cognitive outcomes following exercise in DS, and all of these studies involve acute exercise. This may be, in part, due to the reported difficulty in getting objective measures of physical activity in youth with Down syndrome (Hinckson & Curtis, 2013). In the review they suggest using proxy reports to measure physical activity in children with ID, mainly because of refusal to wear external devices (e.g., pedometers, accelometers, heart rate monitors, etc.). However, there are a few recent studies that examined the the cognitive beenfits of exercise in DS. For example, Ringenbach, Albert, Chen, and Alberts (2014) found that performance on a task involving cognitive planning—the Tower of London—improved after an assisted cycling session. Further, Chen, Ringenbach, Crews, Kulinna, and Amazeen (2015) found an improvement in inhibition following treadmill walking in DS; and more recently (Chen & Ringenbach, 2016) found evidence of a dose-response relationship for exercise intensity and inhibition scores in individuals with DS.

For those with DS, certain barriers have been identified to engaging in physical activity. The most notable are competing family responsibilities, reduced physical or behavioral skills, and a lack of accessible exercise programs (Barr & Shields, 2011). Heller, Hsieh, and Rimmer (2003) suggested that there is a need not only for accessible programs for those with DS, but also a need for increased caregiver's awareness of the importance of exercise for this population. There has also been a call for greater effort to promote physical activity in those with DS because of the known benefits it has for improving general health and avoiding chronic disease (Rimmer et al., 2004) as well as the potential for it to be a catalyst for increased self-esteem and social interaction (Jobling, 2001). It is also important to understand the relationship between exercise and executive function in DS because exercise may be an effective way to improve EF in a population with a general EF deficit.

Taken together, research demonstrates there is an EF deficit that is characteristic of DS, individuals with DS engage in less exercise than those without disabilities, and exercise improves the processes that encompass EF in typically developing children and adults. Thus, research is needed to determine whether increasing exercise in those with DS can lead to improvement in their EF skills. Before attempting an intervention study, however, it is important to know whether exercise is correlated with EF among those with DS–that is, do those with DS who exercise more have better EF? Therefore, the purpose of the current study was to determine whether amount of exercise, or physical activity, relates to EF in participants who have DS. Of course it is possible that participants who are generally higher functioning may engage in more exercise and also have higher EF skills. To address this possibility, we will measure adaptive behavior and examine its influence on the relation between exercise and EF. I hypothesize that there will be a significant positive correlation between exercise and measures of EF even when the influence of level of adaptive functioning is controlled.

In the current study, youth with DS wore a pedometer for one week and their parents completed the Children's Leisure Activities Study Survey (CLASS, Salmon, Telford, & Crawford, 2004), the Behavior Rating Inventory of Executive Function (BRIEF; Gioia, Isquith, Guy, & Kenworthy, 2000), and the Adaptive Behavior Assessment System - Third Edition (ABAS-III, Harrison & Oakland, 2015). The direct (pedometer) and indirect (parent report) measures of physical activity were expected to correlate positively with the BRIEF. Adaptive functioning is expected to mediate this relation, but only partially. After the mediating effect is accounted for, it is expected that there will remain a significant relation between exercise and EF.

2. METHODOLOGY

Design

I used a correlational design to examine the relations between the dependent variable, exercise, the independent variable, EF, and the mediator variable – adaptive functioning. Adaptive functioning was included in the model to clarify the relation between exercise and EF. For example, if exercise is positively related to EF, this could be because those who are higher in adaptive functioning are more active and have higher EF. Conversely, there could be a direct relation between exercise and EF beyond any influence of adaptive functioning level.

Participants

Participants had been previously diagnosed with DS and were ages 6-18 years old. Of the 26 participants, there were 11 males and 15 females, with a mean age of 11.07 (3.84). I recruited participants from the University of Alabama Intellectual Disabilities Participant Registry (UAIDPR). The UAIDPR is a registry used to match researchers with children and adults who have intellectual disabilities. The UAIDPR recruits participants from Alabama, Mississippi, Georgia and Florida for behavioral research. Participants were excluded if their active caregivers had not been their caregivers for more than a year, if their hearing or vision was poor based on parent report, and if English was not their first language. Twenty-six total families participated and 14 of the 26 provided full data for analysis; additionally, 25 of the 26 filled out all of the parent forms, but some were unable to provide pedometer data.

Measures

Adaptive behavior measure. Caregivers of the participants with DS were interviewed using the Adaptive Behavior Assessment System – Third Edition (ABAS-III, Harrison & Oakland, 2015). Specifically, I used the parent form to assess adaptive behavior of our participants. The parent form is designed for children ages 5-21. The ABAS-III assesses the skills that are used in conceptual, social and practical areas of adaptive behavior. The skills assessed from the parent form are communication, community use, functional academics, home living, health and safety, leisure, self-care, self-direction, social, and work. It is important to note the skill "work" is only administered to those who are 17 years old or older, and have a part- or full-time job. It is also not included in the general adaptive composite (GAC). The raw score from the assessment is translated to a scaled score and the scaled scores make the GAC. The parent raters will score each item on a 4-point Likert-type scale with the choices: *is not able*, *never or almost never when needed*, *sometimes when needed*, and *always or almost always when needed*. The test-retest reliability is .88 for the GAC for ages 16-89 (Harrison & Oakland, 2015).

Physical activity measures. Measuring physical activity in participants who have intellectual disabilities is difficult for multiple reasons (e.g., differences in gait, tampering with measurement equipment, etc.). Though there are many limitations with this population, it has been suggested that reports by parents or caregivers are one of the better ways to measure physical activity in individuals with intellectual disabilities (Hinckson & Curtis, 2013). It has also been suggested that no single measure is appropriate to accurately measure physical activity (Chaumeton, Duncan, Duncan, & Strycker, 2011; Welk, Corbin, & Dale, 2000). A study looking at the use of pedometers in adults with DS noted that though there may be differences in gait between typically developing individuals and those with DS, use of a piezoelectric pedometer (also known as an accelerometer pedometer) would minimize the differences in gait as opposed to a spring-levered pedometer (Pitchford & Yun, 2010). Essentially, a piezoelectric pedometer will more accurately assess when participants are taking steps; because of the differences in gait, a spring-levered pedometer may count a step when one was not taken or fail to count a step when one was taken. Pitchford and Yun (2010) also suggested, as other researchers have, the incorporation of multiple methods in assessing physical activity in individuals with Down syndrome, noting the limitations of using pedometers alone.

The current study used both parents' reports as well as a week of pedometer data to achieve a comprehensive and accurate assessment of physical activity. The Omron HJ-720ITC pedometer (a piezoelectric pedometer) was used and parents recorded the number of steps at the end of each day on a tracking-sheet for a seven-day period. The tracking-sheet had a place to provide the number of steps for a day and a space to provide comments about that day as well.

The Children's Leisure Activities Study Survey (CLASS, Salmon, et al., 2004) is a proxy report that was adapted for use in the current study. There are not many physical activity questionnaires that are high in reliability but the CLASS is one of the more reliable physical activity questionnaires currently available for children (Chinapaw, Mokkink, van Poppel, van Mechelen, & Terwee, 2010). The test-retest reliability for the proxy and self-report versions ranged from .62 to .94 (Telford, Salmon, Jolley, & Crawford, 2004). The CLASS requires parents to report their child's physical activity during a typical week. The CLASS is set up as a table with the activity listed in the left column, a yes or no question on whether or not the participant engages in this activity during a typical week in the next column, then how many total minutes for each activity for Monday through Friday as well as Saturday and Sunday in separate columns. The CLASS was modified to include items specific to our culture and population, as previously it had been used with TD children in Australia. In addition to the CLASS, the current study included a similarly designed table recording physical activity over the past two years, with a greater focus on organized recreation activities (sports teams, exercise classes, etc.) The additional table recording the past two years is broken down into school semesters to help parents understand (i.e., spring 2014, fall 2013, and summer 2013). Please see *Appendix* for more details.

Executive function measure.

Behavior Rating Inventory of Executive Function – (BRIEF). The BRIEF (Giola et al., 2000) is a parent proxy report designed to assess children with disorders in EF from ages 5-18. The BRIEF was chosen because it allows us to look at age-based norms via T-scores. The current study used the parent report version of the BRIEF, which uses a 3-point scale for the 86-item questionnaire: *Never*, *Sometimes*, and *Often*. Examples of prompts parents would respond to are: “Has a short attention span,” “Keeps room messy,” and “Gets out of seat at the wrong times.” It is suggested that both parents, or caregivers, be present to compete the questionnaire and it takes approximately 10-15 minutes to complete. The Cronbach’s alpha for the BRIEF parent report ranges from r= .80 to r= .89, with test-retest reliability coefficients ranging from .76 – .85+ with the majority of the scores .80 and above. The BRIEF also has adequate validity (Giola et al. 2000). The BRIEF consists of eight subscales: Inhibit, Shift, Emotional Control, Initiate, Working Memory, Plan and Organization, Organization of Materials, and Monitoring; and three indices: Behavioral Rating Index, Metacognitive Index, and the General Executive Composite (GEC). Parents were given a frame-of-reference in their instructions for filling out the BRIEF; referring to “a typically developing child of the same age” if needed. In the current study BRIEF T-scores were computed and used in the data analysis. T-scores are age-based normative scores with a mean of 50, and a standard deviation of 10. Higher scores on the BRIEF indicate poorer EF.

Procedure

Parents of participants were mailed the modified CLASS, BRIEF, ABAS-III, a background questionnaire, and a pedometer and tracking sheet used to record the daily walking activity for seven days. The caregiver was also given instruction on where to put the pedometer as well as when to record the daily walking activity. Once the pedometer data were collected and the forms were completed, participants mailed back forms and pedometer in the pre-paid postage. If parents did not have questions and did not return the forms for three weeks I would contact them every one to two weeks to answer questions and discuss other logistical details related to the study (i.e., whether or not their child would wear the pedometer, reminding them to send materials back, etc.).

3. RESULTS

Exercise

Pedometer Data. An average steps/day variable was computed for the pedometer data. It was computed by summing the number of steps, for up to five days, and dividing by the number of days that were summed; this procedure is similar to one previously used by Strycker, Duncan, Chaumeton, Duncan, and Toobert (2007). If a participant had seven days of pedometer data (without missing data), the highest and lowest days were removed. For two participants, four days of pedometer data were used to compute the average because there were not five complete days of pedometer data. There was a considerable amount of missing data for the pedometer caused by: complete refusal to wear, partial days, days with 8+ hour car rides, etc. Any missing data were excluded from the analyses, meaning, we did not compute—via multiple imputation or other techniques—participants' missing pedometer data. This decision was made because techniques that compute missing values rely on large samples: those would not have been appropriate give the small sample of the current study. It is also important to note that all variables were normally distributed.

One important question in the study was whether or not participants would wear a pedometer for a seven-day period, and if any pedometers would be lost during the experiment. From our sample, three pedometers were lost—hence the data were lost—and nine participants were unable to wear pedometers. There were no participants that wore the pedometer that were excluded from the analyses. Meaning, participants excluded from data analysis either did not wear the pedometer at all, or lost the pedometer shortly after starting to wear it. From ages 5 to

13 complete pedometer data depended on the participant; however, all participants 13 years and older had complete pedometer data. Anecdotally, the older participants seemed to enjoy wearing the pedometers—many parents commented that they would be purchasing one for their child once the study was completed. The other obstacle we ran into was the amount of time participants needed to collect the pedometer data. Theoretically, it should have only taken a little more than a week or two to finish the parent forms and pedometer data. However, during the study, many participants were very slow at getting started, they would occasionally skip days, and would forget to drop the materials off at the post office once done with the study.

One of the goals the current study was to learn whether or not the average number of steps per day for participants with DS was less than their TD peers. Means and standard deviations for participants are reported in Table 1. As expected, youth with DS report fewer steps than TD youths (approximately 9,000 – 13,000 steps per day) based on information reported from previous studies (Strycker et al, 2007; Tudor-Locke et al. 2011; Beets, Bornstein, Beighle, Cardinal & Morgan, 2010). Additionally, the activity has a similar pattern, such that boys report more steps than girls (see Table 1); however, children did not report significantly more steps per day than adolescents.

Table 1
Means, Standard Deviations (SD), and Confidence Intervals (CI) for the Pedometer Data

	Average Steps Per Day				
	n	Mean	SD	CI	t
Overall	14	5622.46	2352.74	4390.04, 6854.88	
Sex					
Male	4	7945.80	2775.71	5225.65, 10665.94	2.948*
Female	10	4693.13	1438.30	3801.67, 5584.58	
Age[a]					
Children	6	6333.43	3333.57	3666.06, 9000.79	0.978
Adolescent	8	5089.24	1258.66	4217.05, 5961.43	

[a]Children were defined as 5-10 years, adolescents were 11-18 years.
* $p < .05$ (two tailed).

Children's Leisure Activities Study Survey Data. The modified CLASS proxy report was used, and a total score was computed by taking the product of the total activity time per week (in hours), and value of the metabolic equivalence of a task (MET) for the respective activity; the sum of all the time (in hours) X MET products was used to create an exercise total. The MET is a measure of exercise intensity based on oxygen consumption, whereby one MET is equal to resting energy expenditure. The compendium for physical activities (Ainsworth et al., 2011) lists the METs for different activities. Further, vigorous physical activity was of additional interest, so activities were categorized as moderate or vigorous based on their METs. The moderate activities consisted of METs below 5.8, and the vigorous activities had METs of 5.8 and above. The cutoff values were chosen based on a combination of the guidelines posted by Haskell et al. (2007), and 5.8 was the median MET of activities listed on our modified CLASS (i.e., a median split). The same procedure outlined above was used to calculate the total amount of moderate intensity and vigorous intensity physical activity. When calculating CLASS scores, the same MET activity values were used across all participants. If a participant added an activity not already on the CLASS, the corresponding MET was chosen from the compendium of physical activities. There were no statistically significant differences between groups in the amount of total or vigorous activity for sex or age groups reported on the CLASS (see Table 2). However, there was a marginally significant difference in vigorous activity between children and adolescence (*p* = .07) such that children participated in more vigorous activity in a typical week.

Table 2

Means, Standard Deviations (SD), and Confidence Intervals (CI) for the Modified CLASS Data

		All Activity				Vigorous Activity[a]			
	n	Mean	SD	CI	*t*	Mean	SD	CI	*t*
Overall	26	53.38	22.34	44.35, 62.40		29.50	18.06	22.20, 36.79	
Sex									
Male	11	55.88	19.91	42.50, 69.26	0.482	28.21	18.33	15.90, 40.53	0.306
Female	15	51.54	24.49	37.98, 65.10		30.44	18.45	20.23, 40.66	
Age									
Children	14	54.73	21.29	42.44, 67.03	0.328	35.38	18.70	24.58, 46.17	1.881
Adolescence	12	51.79	24.37	36.31, 67.28		22.64	15.26	12.95, 32.34	

[a]Activities with an MET of 5.8 or higher.

Executive Function

General EF, as indicated by the GEC was significantly impaired in comparison to age-based norms. We conducted a series of one-sample T-tests on the mean T-scores of the subscales and indices with a Bonferroni adjusted alpha-level of .0045 per test (.05/11; Table 3), comparing to a normative mean T-score of 50. The subscales that were significantly above the normative mean—indicating poorer EF—were inhibition, shifting, initiating, working memory, planning and organization, and monitoring. The subscales that were not significantly different than the normative mean were emotional control and organization of materials–indicating relative strengths for those with DS. The results of the current study replicated previous research on the EF profile in DS using the BRIEF (Daunhauer et al, 2014; Lee et al., 2015). The percentages of participants in the clinically elevated range (%CE; T-scores ≥ 65) are reported in Table 3. Of particular interest are the pronounced weaknesses in working memory, inhibition, and planning and organization.

Table 3
Percent Clinically Elevated, Means, and T-tests for the BRIEF T-scores

	T-Scores, % CE, and One-Sample T-Tests			
	%CE[a]	Mean	SD	*t*[b]
Inhibit	42.86%	64.85	10.74	7.31*
Shift	21.43%	58.69	10.75	4.28*
Emotional Control	10.71%	50.77	8.24	0.49
Behavioral Rating Index	25.00%	59.19	9.33	5.22*
Initiate	28.57%	59.62	8.04	6.33*
Working Memory	53.57%	66.73	11.26	7.86*
Planning and Organization	53.57%	66.88	14.25	6.27*
Organization of Materials	21.43%	54.46	11.06	2.13
Monitoring	60.71%	68.58	11.42	8.61*
Metacognitive Index	46.43%	65.92	10.43	8.08*
GEC	39.29%	64.20	9.44	7.96*

[a]Percentage of participants in the clinically elevated range (T-scores ≥ 65)
[b]Comparison to normative mean T-score of 50.
*Significant after Bonferroni correction.

Exercise and Executive Function.

Correlations between exercise and EF measures are shown in Table 4. Counter to expectations, the positive correlation between average steps per day and T-scores of the BRIEF was approaching significance, indicating that those who reported more steps also reported poorer EF. Because EF correlated so strongly with age, partial correlations are also reported controlling for the influence of age (Table 5). None of the relationships of interest reached statistical significance for the partial correlations. After reviewing the correlations of the key variables, there was no relationship found between EF and adaptive function. Thus, the main hypothesis that exercise is related to EF is not supported, and no mediation analysis was computed.

Table 4
Bivariate Pearson Correlation Among Key Variables

		1	2	3	4	5	6
1	Age						
2	Average Steps Per Day (n=14)	-0.279					
3	CLASS Total (n=26)	0.015	0.410				
4	CLASS Moderate (n=26)	0.159	0.146	0.484*			
5	CLASS Vigorous (n=26)	-0.370	0.456	0.652**	-0.075		
6	General Adaptive Composite (n=25)	-0.112	-0.155	0.159	0.230	0.141	
7	General EF (n=26)	-0.502*	0.406	-0.179	0.008	-0.060	-0.252

* *p* <.05 (two-tailed).
** *p* <.01 (two-tailed).

Table 5
Partial Correlations Among Key Variables Controlling for Age

		1	2	3	4	5
1	Average Steps Per Day (n=14)					
2	CLASS Total (n=26)	0.432				
3	CLASS Moderate (n=26)	0.201	0.488*			
4	CLASS Vigorous (n=26)	0.396	0.708**	-0.017		
5	General Adaptive Composite (n=25)	-0.196	0.162	0.253	0.108	
6	General EF (n=26)	0.320	-0.198	0.103	-0.306	-0.359

* *p* <.05 (two-tailed).
** *p* <.01 (two-tailed).

4. DISCUSSION

The main purpose of the current study was to determine if there is a relationship between physical activity and EF in DS. We expected that those who reported higher amounts of physical activity would have better EF. There was no significant correlation between scores on the CLASS and EF. Further, the mediating variable—adaptive functioning—was not related to either measure of physical activity or to EF. Nevertheless, some interesting descriptive findings emerge from the study.

Exercise and Executive Function

Importantly, the EF profile replicates findings from previous studies in youth with DS using the BRIEF (Daunhauer et al, 2014; Lee et al., 2015). Youth with DS have strengths in emotional control and organization of materials, and weaknesses across the other subscales. The main analysis in the present study was expected to reveal a significant positive relationship between exercise and EF after controlling for the influence of adaptive behavior, but a nonsignificant trend in the opposite direction was found in average steps per day (i.e., more steps per day were related to poorer EF). The initial take-away may be that poorer EF, as measured by the BRIEF, is associated with increased average steps per day. However, previous research suggests that exercise is beneficial for executive function in children (Best, 2010; Davis et al., 2011; Tomprowski et al., 2008); there are even acute benefits found immediately after walking in a DS population (Chen, Ringenbach, Crews, Kulinna, & Amazeen, 2015). Thus, the following are several alternative explanations for our findings.

First, the BRIEF may assess slightly different underlying processes than traditional tasks used to assess EF (e.g., Wisconsin Card Sort Task, Stroop task, Rule Shift card task, reverse digit span, Tower of London, etc.). Toplak, West, and Stanovich (2013) reviewed 13 studies that used the BRIEF along with performance-based measures of EF and concluded that the association between the two was very weak. Only 19% of the correlations that were reported between performance-based measures of EF and the BRIEF were significant. The mean and median correlation coefficients were .15 and .18 respectively, and it is likely that many nonsignificant correlations were not published. Toplak and colleagues (2013) proposed that performance based measures of EF provide an indication of processing efficiency, whereas rating-forms provide a clearer indication of individual goal pursuit. This would imply that the previous research—finding that exercise is beneficial for EF—referred to performance-based measures because few, if any, studies investigating exercise and EF used rating forms (i.e., the BRIEF). In sum, it may be that the current study found that average steps per day might be related to poorer goal pursuit, or behavioral EF. Given that approximately half of the sample had usable pedometer data, and the correlation was not significant, this conclusion is very speculative.

Second, the type of physical activity may make a large difference. Average steps per day is not a measure of vigorous physical activity, and cannot be thought of as a proxy for all physical activity. Several studies on exercise and cognition in children include interventions that involve *moderate to vigorous* physical activity (see Best 2010; and Strong et al., 2005). Recently Castelli, Hillman, Hirsch, Hirsch, and Drollette (2011) suggested that there is a dose-response relationship in TD youths, with more cognitive benefits associated with increasingly vigorous activity. Interestingly, average steps per day and vigorous activity reported in the CLASS trended with the GEC in opposite directions (see Table 4). This indicates they are either measuring different constructs, or capturing separate variance related to one construct (i.e., physical activity). Given the theoretical importance of vigorous activity (i.e., more vigorous activity is cognitively beneficial for children and adolescents, Castelli, 2011) its relationship with the GEC is encouraging. It is important to note, the conclusions we draw from the average steps per day are limited by the small sample. Unfortunately, several participants reported having difficulties with keeping the pedometers on throughout the day.

Exercise

The current study provides novel information on the physical activity levels, especially in the form of average steps per day, in youth with DS. Youth with DS reported lower average steps per day than what is typical for their age group. However, the pattern of average steps per day is the same in DS as it is in TD youths, such that boys reported more steps than girls. There was also a nonsignificant trend for younger children to record more steps per day than adolescents. These descriptive findings are especially important because the current study is the first recording average steps per day in youth with DS. Tudor-Locke, et. al (2011) reported the average steps per day for TD boys and girls ages 6-11 as 13,00 and 12,000, respectively; boys and girls ages 12-19 reported 11,000 and 9,000 steps per day respectively. Participants in the current study reported taking fewer steps per day, which is consistent with previous research reporting lower activity levels in youth with ID (see Hinkerson & Curtis, 2013).

A review paper by Hinkerson and Curtis (2013) noted the difficulty of obtaining objective measures of exercise in youth with ID—we encountered some difficulties using pedometers. The older participants gave complete pedometer data, and some of the younger participants did not. Additionally, no older participants (i.e., ≥ 13 years old) reported losing the pedometer. Many parents commented that their child enjoyed wearing the pedometer, so much so that many parents of older participants asked where they could buy a similar one. Our difficulties with younger participants are consistent with previous accounts of difficulties related to using objective measures (see Hinkerson & Curtis, 2013) in ID populations; however, we tentatively suggest the age of 13 may be an approximate cutoff where pedometers become a viable measure of physical activity in DS.

Limitations

It is important to address a few limitations in the current study. The most notable is the small sample size in the study. Unfortunately, we are missing data because many participants had issues with the pedometer—limiting the conclusions we can draw from the pedometer data. Another limitation is the potential for bias in parent/caregiver reports of their children's adaptive behavior, executive function, and physical activity. Although the measures are designed to assess trait-like behaviors rather than state-like behaviors, parents may have been influenced by their child's behavior that day, or shortly before filling out the forms. The physical activity questionnaire is also limited by parents' ability to report what their child does throughout the day, including activities like: physical education classes at school. In the current study, parent-omitted items were assumed to be true omissions, but the parents may have forgotten to report some activities.

Future Directions

Our findings indicate that more work needs to be done to improve the measurement of exercise in DS (e.g., slightly altering the CLASS to reduce parental omissions, and fine-tuning or finding a different objective measure of exercise in DS). Parental omissions may be reduced by asking parents to include hour by hour detail on what their children do during a typical day: asking parents to include time spent sitting, traveling to school, sleeping, walking the dog, etc., and ensuring it adds up to close to 24 hours. For increased pedometer participation (i.e., participants correctly wearing them), it may help to have a smaller and/or more secure pedometer to reduce the number of children not wanting to wear it or losing it. An alternative objective measure of physical activity may be the use of a Fitbit-like device. Pending acceptable validity and reliability, a Fitbit-like device may be a great option in the future. Anecdotally, many parents who had issues collecting data with the pedometer offered to have their child wear their Fitbit because their child enjoyed wearing it. Future studies measuring EF would benefit from having a rating form as well as performance-based measures of EF because they are theoretically nuanced. Additionally, future studies will be needed to better understand if there is a dose-response relationship between exercise and EF in youth with DS (e.g., such that vigorous activity is more beneficial for EF than less vigorous activity). Most of all, this current study highlights the need for more research to better understand the complex relationship between exercise and EF in DS.

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APPENDIX A

During a typical WEEK what activities does your child **usually** do?	Does your child usually do this activity?	**Monday - Friday**		**Saturday - Sunday**	
		How Many times **Monday-Friday**	Total hours/minutes **Monday-Friday**	How Many times **Saturday & Sunday**	Total hours/minutes **Saturday & Sunday**
E.g. Bike Riding	No1 **Yes2**	**2**	1hr 10mins	1	20mins
Aerobics class	No1 Yes2				
Baseball / Softball	No1 Yes2				
Basketball	No1 Yes2				
Bike riding	No1 Yes2				
Bounce on trampoline	No1 Yes2				
Dance	No1 Yes2				
Gymnastics	No1 Yes2				
Household chores	No1 Yes2				
Jogging or running	No1 Yes2				
Jumping rope	No1 Yes2				
Physical education class	No1 Yes2				
Play on playground	No1 Yes2				

equipment					
Play with pets	No_1 Yes_2				
Roller blading	No_1 Yes_2				
Scooter	No_1 Yes_2				
Skateboarding	No_1 Yes_2				
Soccer	No_1 Yes_2				
Swimming for fun	No_1 Yes_2				
Swimming laps	No_1 Yes_2				
Tennis	No_1 Yes_2				

During a typical WEEK what activities does your child **usually** do?	Does your child usually do this activity?	**Monday - Friday**		**Saturday - Sunday**	
		How Many times **Monday-Friday**	Total hours/minutes **Monday-Friday**	How Many times **Saturday & Sunday**	Total hours/minutes **Saturday & Sunday**
Tennis	No_1 Yes_2				
Volleyball	No_1 Yes_2				
Walk for exercise	No_1 Yes_2				
Walk the dog	No_1 Yes_2				
Travel by walking to school (to and from school = 2 times)	No_1 Yes_2				
Travel by cycling to school (to and from	No_1 Yes_2				

school = 2 times)					
Other (please state below)	No1 Yes2				
	No1 Yes2				
	No1 Yes2				
	No1 Yes2				
	No1 Yes2				
	No1 Yes2				
	No1 Yes2				
	No1 Yes2				
	No1 Yes2				
	No1 Yes2				
	No1 Yes2				
	No1 Yes2				
	No1 Yes2				
	No1 Yes2				

The next questions will ask about **organized physical activity** from past **two years**. It is organized by the school's calendar-year. Fall is from September-December, Spring is from Janurary to May, and Summer if from June-August

In the **SPRING of 2014** what physical activities did your child particiapte in?	How many **days** per week?	**Total** hours/minutes per week	Additional information

E.g. Tennis lessons	3	3hr 30mins	Only for an 8-week period
In the **FALL of 2013** what physical activities did your child particiapte in?	How many **days** per week?	**Total** hours/minutes per week	Additional information
In the **SUMMER of 2013** what physical activities did your child particiapte in?	How many **days** per week?	**Total** hours/minutes per week	Additional information

Continued on next page…

In the **SPRING of 2013** what physical activities did your child particiapte in?	How many **days** per week?	**Total** hours/minutes per week	Additional information
In the **FALL of 2012** what physical activities did your child particiapte in?	How many **days** per week?	**Total** hours/minutes per week	Additional information
In the **SUMMER of 2012** what physical activities did your child particiapte in?	How many **days** per week?	**Total** hours/minutes per week	Additional information

In the **SPRING of 2012** what physical activities did your child particiapte in?	How many **days** per week?	**Total** hours/minutes per week	Additional information

APPENDIX B

THE UNIVERSITY OF ALABAMA® | Office of the Vice President for Research & Economic Development
Institutional Review Board for the Protection of Human Subjects

July 15, 2016

Andrew Tungate
Department of Psychology
College of Arts & Sciences
Box#: 870348

Re: IRB Protocol 14-013-R2
"Exercise and Executive Function in Down Syndrome"

Dear Mr. Tungate:

The University of Alabama Non-Medical IRB recently met to consider your renewal application. The IRB voted to approve your protocol for a one year period.

Your application will expire on July 14, 2017. If your research will continue beyond this date, complete the renewal portions of the IRB Renewal Application. If you need to modify the study, please submit the Modification of an Approved Protocol Form. **Changes in this study cannot be initiated without IRB approval, except when necessary to eliminate apparent immediate hazards to participants.** When the study closes, please complete the Request for Study Closure.

Please use reproductions of the IRB approved stamped consent/assent forms to obtain consent from your participants.

Should you need to submit any further correspondence regarding this application, please include the above application number.

Good luck with your research.

Sincerely,

[redacted]

Stuart Usdan, PhD
Chair, Non-Medical Institutional Review Board

358 Rose Administration Building | Box 870127 | Tuscaloosa, AL [illegible]
205-348-8461 | Fax 205-348-7189 | Toll Free [illegible]

IRB Project #:14-013 -R2

UNIVERSITY OF ALABAMA INSTITUTIONAL REVIEW BOARD FOR THE PROTECTION OF HUMAN SUBJECTS
REQUEST FOR APPROVAL OF RESEARCH INVOLVING HUMAN SUBJECTS

I. Identifying information

	Principal Investigator	Second Investigator	Third Investigator
Name:	Andrew Tungate	Dr. Frances Conners	Gayle Graham Faught
Department:	Psychology	Psychology	Psychology
College:	A&S	A&S	A&S
University:	UA	UA	UA
Address:	Box 870348	Box 870348	Box 870348
Telephone:	205-348-4253	348-7913	205-233-0124
FAX:	348-8648	348-8648	348-8648
E-mail:	atungate@crimson.ua.edu	fconners@bama.ua.edu	eggraham@crimson.ua.edu

Title of Research Project: Exercise and executive function in Down syndrome
Date Printed: 06/27/16 Funding Source: The PI (Andrew Tungate)
Type of Proposal: __New ____Revision _X_Renewal ___Completed ___Exempt

Attach a renewal application

Attach a continuing review of studies form

Please enter the original IRB # at the top of the page

UA faculty or staff member signature: [redacted]

II. NOTIFICATION OF IRB ACTION (to be completed by IRB):
Type of Review: __X__ Full board _____ Expedited

IRB Action:
___Rejected Date:__________
___Tabled Pending Revisions Date:__________
___Approved Pending Revisions Date:__________
X Approved—this proposal complies with University and federal regulations for the protection of human subjects.
Approval is effective until the following date: 7/14/2017

Items approved:
X Research protocol: dated
X Informed consent: dated
__ Recruitment materials: dated
X Other: assent dated

Approval signature ______________________________Date ________________

Informed Consent for a Research Study

You and your child are invited to take part in a research study called "Exercise and executive function in Down syndrome." The study is being done by Andrew Tungate for his master's thesis, under the supervision of Dr. Fran Conners, a professor at the University of Alabama.

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WHAT IS THE STUDY FOR?

This study will find out if children with Down syndrome who exercise more have higher cognitive functioning. If they do, we might want to see if exercising more actually helps your memory and thinking. Research has already shown this for typically developing children and adults, but not for those with Down syndrome.

WHAT WILL WE DO?

(1) During a typical week that you choose, your child will wear a lightweight pedometer each day and you will write down the pedometer count at the end of each day. The pedometer will measure the number of steps your child takes each day.

(2) Next, a researcher will come to your home (or you may choose to come to the UA campus) for an hour appointment.
   - You will fill out four questionnaires about your child's physical activity, everyday memory and planning, everyday life skills, and basic background (45 min).
   - Your child will do 5 gamelike cognitive tasks - matching pictures, remembering lists, following rules, saying the opposite, and doing the opposite (45 min, with breaks). The researcher will help your child with these tasks while you are filling out the questionnaires.

WHAT INFORMATION WILL YOU KEEP?

We will keep your records from the pedometer, your answers from the questionnaires, and results from your child's cognitive tasks.

ARE THERE ANY RISKS OR BENEFITS TO ME OR MY FAMILY?

There are no special risks in this study. If your child gets bored or frustrated during the cognitive tasks we will take a break. There are no costs to you or your child for being in the study, other than your time and effort. General benefits of participating in the study include the satisfaction of helping researchers learn more about exercise and cognition in individuals with Down syndrome.

WILL WE GET ANYTHING FOR OUR TIME AND EFFORT?

Yes, you will receive either a $35 gift card if you travel to campus for testing or a $15 gift card if testing is done in your home as a token of thanks for your participation. Additionally, your child will receive a ribbon for completing the week with the pedometer and a small prize after completing the cognitive tasks.

HOW WILL OUR CONFIDENTIALITY AND PRIVACY BE PROTECTED?

We will not give information about you or your child to anyone else. We will store your answers and your child's task scores on a private, secure computer that is only used by the researchers. We will use code numbers instead of names for each participant and we will keep the code list locked in a file cabinet.

UNIVERSITY OF ALABAMA IRB
CONSENT FORM APPROVED: 7/15/16
EXPIRATION DATE: 7/14/2017

WHAT OUR RIGHTS AS PERSONS IN A RESEARCH STUDY?

Being in this study is your free choice. Even if you start the study, you are free to change your mind and stop at any time.

WHOM SHOULD I CONTACT IF I HAVE QUESTIONS?

- For questions or comments about the study, call or email Andrew Tungage (205-348-4253 or atungate@crimson.ua.edu) or Dr. Fran Conners (205-348-7913 or fconners@as.ua.edu).
- For questions about your rights as a person in a research study, call Ms. Tanta Myles, The University of Alabama Research Compliance Officer, at 205-348-8461 or toll-free at 1-877-820-3066.
- For suggestions, complaints or concerns:
    - Call Ms. Tanta Myles at 205-348-8461 or toll-free at 1-877-820-3066
    - Use the IRB Outreach Website at http://osp.ua.edu/site/PRCO_Welcome.html
    - Email participantoutreach@bama.ua.edu

If you give permission for your child to participate, please complete the part below.

I understand the purpose of the study and what I and my child will be asked to do. I agree to complete my part of the study and I give permission for my child to be in the study.

Child's full name (please print) ____________________

Parent/Guardian's full name (please print) ____________________

Parent/Guardian's signature ____________________
(Must be a parent/guardian who signs for school/medical purposes)

Parent/Guardian's phone number ____________________

Parent/Guardian's email ____________________

Today's date ____/____/____ Child's birth date ____/____/____

THANK YOU!

Would you and/or your child like to be contacted to help in future research studies? ____ yes ____ no

UNIVERSITY OF ALABAMA IRB
CONSENT FORM APPROVED: 7/15/16
EXPIRATION DATE: 7/14/2017

Appendix B
Protocol for Obtaining Child's Assent

Hi. My name is _______, and I work at the University of Alabama. I study memory and thinking. I am trying to find out if moving around and exercise is related to your memory and thinking. If it is, we might want to see if exercising more actually helps your memory and thinking.

Your parent/guardian said it would be okay for you to help me with my study. Actually, you have already helped me by wearing the pedometer on your waist. This told me how many steps you took each day – in other words, how much you moved around. Today I want you to do some memory and thinking games, if you want to. There are 5 of these games and each one is only a few minutes long. We will probably finish them all today.

The most important thing about these games is that you try your best. Trying your best is more important than getting the right answers.

If you say OK now, but change your mind later and don't want to do the games, just let me know and we'll stop. It's your choice to do this and you can always say no.

So is it ok if we get started?

Child's Name ____

Answer (circle) YES NO Date / / / ____________

(Staple to Informed Consent Form)