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Can activities using virtual reality improve motor function in children with Down Syndrome?

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A SELECTIVE EVIDENCE BASED MEDICINE REVIEW

In Partial Fulfillment of the Requirements For

The Degree of Master of Science

In

Health Sciences - Physician Assistant

Department of Physician Assistant Studies Philadelphia College of Osteopathic Medicine Philadelphia, Pennsylvania

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ABSTRACT

OBJECTIVE: The objective of this selective EBM review is to determine whether or not activities using virtual reality improve motor function in children with Down Syndrome.

STUDY DESIGN: A review of three randomized controlled trials (RCTs) published in English, one of which is also published in Spanish, between the years of 2010-2018.

DATA SOURCES: Three peer-reviewed journal articles were found using PubMed and CINAHL Plus. Articles were selected based on their relevance to the question and whether the outcome measured was patient-oriented.

OUTCOMES MEASURED: The articles analyze motor function in children with Down Syndrome through the Bruininks-Oseretsky Test of Motor Proficiency-Second Edition (BOT-2) in two of the studies assessing motor proficiency, and the Test of Gross Development (TGMD-2) in one of the studies evaluating locomotor and object control skills.

RESULTS: Wuang et al. revealed that participants in the VRWii group utilizing Wii gaming technology had a significant greater pre-post change on motor proficiency compared to those that received only standard occupational therapy with $p \le 0.0001$. Lin et al. denotes significant improvement in motor proficiency following an exercise program consisting of both a treadmill and VR-based activity using Wii Sports with a p-value of 0.01. Gomez et al. found those who utilized a Wii Balance Board had significant improved postural control and thus improved motor skills compared to the control group with a p = 0.002.

CONCLUSIONS: This review provides evidence from three separate studies that incorporating activities using virtual reality can improve the motor function in children with Down Syndrome. None of the studies were able to strictly control activities at home during the time of the trial. Future studies should include activity diaries to closely monitor activities outside of the study as that could impact results, as well as further evaluate whether virtual-based activities could serve as a sole therapy or as an adjunct to the standard therapies already in use today.

KEY WORDS: Virtual Reality, Down Syndrome

INTRODUCTION

Down Syndrome (DS) is the most common chromosomal disorder that occurs at chromosome 21 in which there could be a third chromosome 21 in each cell, a translocation at that chromosome, or an extra chromosome in just some body cells. DS occurs in about 1/800 births and is prevalent in all races.¹ DS is a leading cause of intellectual disability and is characterized by orthopedic, cardiovascular, musculoskeletal and perceptual impairments, in addition to limitations in adaptive behavior. A child with DS has physiological and anatomical characteristics including hypotonia, joint hypermobility, and sensory deficits that contribute to motor developmental delays up to two years later than a child with typical development.² These limitations lead to delays in motor milestones relative to age-matched typically developing children.¹

The exact cause of DS is unknown; however, this genetic anomaly has been associated with risk factors such as advanced maternal age. Parents often know prior to birth whether their child will have DS through prenatal methods such as amniocentesis, chorionic villus sampling, and ultrasound. The sensorimotor difficulties individuals with DS face lead to higher sedentary rates and lower levels of physical activity, ultimately impacting their overall health and leading to a poorer quality of life in their future.³ It is imperative to address the development of muscle strength and agility early on in children with DS to avoid the detrimental health repercussions that could result from lack of targeted therapies. Muscle function is pertinent to daily activities and work-related skills and greatly influences the general health of an individual as lack of physical activity can lead to obesity, cardiovascular disease, osteoporosis, among others.

While there is not an exact estimate of number of health care visits each year for an individual with DS, these children are at higher risk to experience an abundance of health

problems that influence the amount of health care visits.⁴ Reports show incremental out-ofpocket medical costs as high as \$18,248 for patients with DS and \$230,043 for third-party payers between birth and age 18.⁵ By the time a child with DS is four years old, their medical costs are about 12 times higher than a healthy child of the same age.⁴ Early interventional therapies are pertinent to their development, especially the typical methods used to improve motor function, which include: physical therapy or occupational therapies such as sensory integrative (SI) therapy, perceptual motor (PM) approach, and neurodevelopmental treatment (NDT).¹

Different strategies have been used to improve motor skills ranging from specific strategies for fundamental skills to whole-body vibration training. However, the general overall problem observed of children with DS is the lack of motivation and essential commitment to the intervention to have successful improvement in their motor function.² Children with disabilities often struggle with therapies that are repetitive in nature; typical therapies, such as PT and OT, commonly encompass such repetition, therefore decreasing a child's overall motivation. The introduction of incorporating commercially available VR-based systems as an effective and motivating therapy for children with DS would expand accessibility as they are easily available, affordable, and can be used in the home. The inherent fun experience virtual reality has to offer is attractive to children and may favor adherence early on.² This paper evaluates three randomized controlled trials comparing the efficacy of activities incorporating virtual reality-based intervention for improving motor proficiency and motor skills in children with DS.

OBJECTIVE

The objective of this systematic review is to determine whether or not activities using virtual reality improve motor function in children with Down Syndrome.

METHODS

The three randomized controlled trials selected for this review focus on children with Down Syndrome under the age of 18. The studies were all published in peer-reviewed journals and were found using PubMed and CINAHL Plus using the key words "virtual reality" and "Down Syndrome". All three articles are published in English - one of which is published in Spanish as well - and were selected based on relevance to the clinical question and if they included patient-oriented outcomes. Inclusion search criteria included studies that were clinical trials, randomized controlled trials, publication dates within the last 10 years, population of humans, and written in English. Exclusion criteria for the individual study characteristics are provided in Table 1. The statistics reported in the studies and utilized for this review were mean change from baseline, Cohen *d* value, and p-value. While all of the studies focus on activities that incorporate virtual reality, two of the studies utilize the Wii gaming system solely as the intervention, while one incorporates the Wii into an exercise program that also includes a treadmill exercise.

Wuang et al. compares a group of children with DS that participated in Wii Sports to a group of children with DS that received standard occupational therapy, each for two 60-minute sessions per week for 24 weeks, measuring the outcome of motor proficiency.¹ Lin et al. compares a group of children with DS who partake in an exercise program that consists of a five-minute treadmill exercise and one twenty-minute Wii Sports game three times per week for six weeks to a group of children with DS that does not participate in the exercise program observing muscle strength and agility.³ Gomez et al. compares a group of children with DS that utilizes a Wii Fit software and Wii Balance Board Group two times per week for five weeks to a group of

children with DS that does not perform any intervention and continues with normal daily

activities, observing the outcome of motor development and postural control.²

Study	Туре	#	Age	Inclusion	Exclusion Criteria	W/D	Interventions
Wuang ¹ (2010)	RCT	Pts 105	(Yrs) 7-12	Criteria -Aged 7-12 -Dx of DS by board-certified physicians	-Children who carried coexisting autism, CP, blindness, and deafness -Children with previous hx of neurological	0	VR Wii gaming technology – Wii Sports for 60 min sessions 2 days a week for 24 weeks
Lin ³ (2012)	RCT	92	13-18	-Age13-18 & current high school enrollment -Dx of DS by board-certified physician -Able to follow simple instructions -Written consent	disorders - Subjects with associated cardiovascular conditions, blindness, deafness, or previous neurological impairment -Subject who received any physical or occupational tx in year preceding study	0	Exercise Training program of 5-min treadmill exercise and one 20-min VR-based activity (Wii Sports games) 3 times a week for 6 weeks
Gomez ² (2018)	RCT	16	6-12	 Dx of DS Signed authorization by their guardian Able to comply with orders assigned by researchers 	-Dx of heart diseases -Dx of another disability -Failed to meet 85% of planned sessions	0	VR-based intervention Wii Fit software with Wii Balance Board Group 20-minute sessions 2 days a week for 5 weeks

 Table 1. Demographics and Characteristics of Included Studies

OUTCOMES MEASURED

The outcomes measured in the studies focus on motor proficiency and motor skills. Both Wuang et al. and Lin et al. measured motor proficiency using the Bruininks-Oseretsky Test of Motor Proficiency-Second Edition (BOT-2). The BOT-2 assesses proficiency in four motor area composites that are further subdivided and scored by occupational therapists blinded to child status, eventually combined to generate a total motor composite score.¹ A higher score suggests better motor abilities. First, the fine manual control composite is subdivided into fine motor precision and fine motor integration. Second, the manual coordination composite is subdivided into manual dexterity and upper-limb coordination. Third, the body coordination composite is subdivided into bilateral coordination and balance. Lastly, the strength and agility composite is subdivided into running speed and agility and strength. Lin et al. solely focused on the strength and agility composite. In the third study, Gomez et al. measured the gross motor development through the Test of Gross Development (TGMD-2), which is also a scoring system administered by trained professionals that evaluates both locomotor skills and object control skills to determine the deficits in the motor development of the children.²

RESULTS

Wuang et al. conducted a randomized controlled trial with two interventions of either standard occupational therapy (SOT) or virtual reality using Wii gaming technology (VRWii) to compare the effects on the children with DS.¹ Their study consisted of 105 children with DS randomly assigned to one of the two groups using a computer-generated random table with 52 children in the VRWii group and 53 children in the SOT group.¹ All children who entered the trial were accounted for throughout the entirety of the study. The Wii works by using a wireless controller that interacts with the player through a motion detection system and has an avatar representation in the video. This study specifically used the publicly available Wii Sports software.¹ The SOT incorporated a SI approach which included activities such as linear and circular swinging, tactile-perception, bilateral integration and sequencing, and equilibrium reactions, a NDT approach focusing on postural control and optimal movement patterns which included activities such as developmental movement patterns, walking, strengthening of antigravity muscles, and fine motor skills, as well as a PM approach which included fine and gross motor training.¹

Each intervention was administered under the guidance of two therapists who were randomly assigned to each child to conduct the intervention.¹ Additionally, two pediatric occupational therapists who were blind to each child's group status administered the BOT-2 before and after the intervention.¹ The VRWii group significantly outperformed the SOT group on the three BOT-2 subtests: fine motor integration, upper-limb coordination, and running speed and agility.¹ The mean change from baseline and the Cohen *d* values, which reflect the effect size for the pre-post comparisons across the two intervention groups, are displayed in Table 2. While the VRWii therapy produced the larger effect sizes, with the exception of manual dexterity, measured by BOT-2 compared to the SOT therapies, both interventions illustrated significant improvement. For example, while the SOT group had a positive mean change from baseline of 1.64, the VRWii group had a mean change of baseline of 2.42.¹ The treatment effect size is depicted through the Cohen *d* value, with a medium effect size of Cohen $d \ge 0.5 < 0.8$, and a large effect size of Cohen $d \ge 0.8$. Between groups, there was a statistically significant difference with a p-value $\le 0.0001.^1$

	VRWii Group SOT Group							
BOT-2	Pre-	Post-	Mean change	Cohen d,	Pre-	Post-	Mean change	Cohen d,
	test	test	from baseline	Effect size	test	test	from baseline	Effect size
	mean	mean			mean	mean		
Fine Motor	8.23	10.65	2.42	2.47,	8.28	9.92	1.64	1.56,
Integration				Large				Large
Upper-limb	7.96	10.62	2.66	2.33,	8.11	9.32	1.21	1.08,
Coordination				Large				Large
Running	7.38	10.12	2.74	2.56,	7.47	9.36	1.89	1.73,
speed and				Large				Large
agility				_				_
Manual	5.94	6.44	0.50	0.5,	6.08	6.68	0.60	0.59,
Dexterity				Medium				Medium
Significant difference between groups with p-value ≤ 0.0001								

Table 2. VRWii Group compared to SOT Group Results¹

Lin et al. carried out a randomized controlled trial with 92 adolescents diagnosed with DS randomized to either an experimental group, who underwent a strength and agility training program, or a control group, who did not receive any intervention and continued with their daily life, to determine the effects on children with DS.³ Further inclusion and exclusion criteria for the study can be found in Table 1. All participants were successfully accounted for throughout the entirety of the study. The exercise training program lasted six weeks and included a five-minute treadmill exercise, a ten-minute break, and then a twenty-minute Wii Sports activity.³ The treadmill used was the Sunpro Treadmill 005 and active stretching was allowed prior to walking, with speed and instrument elevation determined in advance by a therapist.³ The participants were allowed to select their preferred VR activity out of 15 pre-selected Wii Sports items.³ The participant used the system by themselves for the first six sessions, worked with a therapist or staff member for sessions seven through ten, and played with two or three other participants for sessions eleven through eighteen.³ Two pediatric occupational therapists blinded to the child group status administered the measurements for the BOT-2 tests.³

Lin et al. focused on the strength and agility composite of the BOT-2 to measure results.³ Prior to the intervention, both the experimental and control groups had a mean total agility score of 11.³ For the group that received the training program, the mean score increased to 16, while the mean score of the control group decreased to 10.³ The mean change of baseline therefore when looking at the total agility score for the experimental group is 5, while the control group is -1.³ These results have a statistical significance of p-value = 0.01, and the treatment effect was considered large with a Cohen *d* value of 0.8.³

		Exercise G	roup	Control Group			
BOT-2	Pre-test Post-test		Mean change	Pre-test	Post-test	Mean change	
	mean	mean	from baseline	Mean	mean	from baseline	
Total Agility	11	16	5	11	10	-1	
Score							
p-value = 0.01, Cohen <i>d</i> value = 0.8							

Table 3. Exercise Group compared to Control Group Results³

Gomez et al. carried out a randomized controlled trial in which 16 children with DS were randomly assigned to either an experimental group (WBBG), who participated in an activity with a Wii Balance Board for twenty-minute sessions two times per week for five weeks, or to a control group (CG), who did not perform an intervention, to determine the effect of VR-based intervention on motor development.² Detailed inclusion and exclusion criteria for the study can be found in Table 1. All participants were successfully accounted for throughout the entirety of the study. The games selected utilized the Wii Fit software with the Wii Balance Board.²

A significant increase measured with TGMD-2 was observed in the WBBG, while no significant changes were observed in the CG. The mean change of baseline for the WBBG was 8.67 while the CG mean change of baseline was -0.72.² Additionally, based on a significance of p < 0.05, the WBBG had a p-value = 0.002 which was significant, while the CG p-value = 0.60 which was not significant.²

WBBG (n=9) CG (n=7) Mean change Mean change **Pre-test** Post-test **Pre-test** Post-test of baseline of baseline mean mean mean mean TGMD-2 63.00 71.67 8.67 63.86 63.14 -0.72 **WBBG: p-value** 0.002*, **CG: p-value** 0.60

Table 4. WBBG compared to CG Results²

Tolerability was not discussed and there were no adverse effects or injuries sustained that were noted in any of the studies.

DISCUSSION

Down Syndrome is a leading cause of intellectual disability that has multiple adverse effects on the motor function development of children. This review highlights three randomized controlled trials that evaluate the effectiveness of activities using virtual reality on motor function for children with DS. Each study demonstrated statistically significant improvements following their intervention with p-values ≤ 0.0001 , 0.01, and 0.002 in Wuang et al., Lin et al. and Gomez et al., respectively.^{1,3,2}

VR-based activities that include the Nintendo Wii have been used for therapeutic purposes, revealing an effect on balance, motor function, energy efficiency, and postural control on people with various conditions, including: cerebral palsy, burn injuries, strokes, cancer, amputations, Parkinson's disease, or spinal cord injuries.² Children with disabilities, especially those with intellectual disabilities, have struggled with the typical PT and OT therapies due to the inherent repetitive nature and the resulting lack of motivation. The Wii program and the immediate positive feedback it has to offer breaks this repetition. Therefore, interventions incorporating VR that are child-centered and offer a greater element of fun could favor adherence and motivation, leading to significant improvement of motor function and hypotonia, while reducing risk of obesity. If not addressed early on, these factors can lead to further health complications later in life.²

The above-mentioned studies provide evidence that participating in activities that incorporate VR can improve the motor function in children with DS. While these studies do not prove that VR-based activities should be used as a sole therapy nor as a replacement to PT and OT, it could be used as an additional modality or for those that may have limited access. The Wii offers promise as an adjunctive therapy to traditional methods, as it is widely available, affordable, easy to use at home, and does not require insurance or trained professionals. Additionally, the ability to continue using VR-based programs, like the Wii, in a home environment allows children to sustain effective physical activity for a longer period of time compared to PT or OT sessions, which are time-limited. The Wii can offer a strong clinical applicability for children as it instantly captures and reproduces their movements via an infrared light sensor. This immediate feedback generates positive reinforcement for a child and successfully facilitates the therapy, especially for those with intellectual limitations.¹ While the standard therapies remain to be beneficial, the addition of a VR-based program can further advance the motor skills and improve the quality of life for individuals with DS.

These studies are not without limitations. All three studies discussed admit they were unable to control the activities performed by the participating children outside of the intervention in their home environments. Wuang et al. suggests that future studies could have guardians log the intensity and frequency of physical activities the children perform while home.¹ Furthermore, the generalizability of the results should be done so with vigilance. Lin et al. included participants that were considered to have moderate intellectual disability, therefore generalizing the results for children with more severe intellectual disability should be done so cautiously.³ Also, Gomez et al. used a small convenient sample of only sixteen participants, nine in the WBBG and seven in the CG, so the results have a limited generalizability to a larger population.² Finally, they all lacked long-term follow up; therefore, while each study was able to illustrate improvement following their intervention, they cannot prove lasting benefits. A child with DS potentially can acquire muscle strength within four to eight weeks, so a six-week program may be sufficient to determine appropriate efficacy, but not long-term effect.³ While these studies generate promising results, they were comprised of limited sample sizes and a lack of control over outside activities with no long-term follow up. In addition to improving the above-mentioned deficiencies, future studies that both directly compare and combine current PT and OT therapies with VR-based activities would further be able to deduce the effectiveness of VR-based activities as a therapy for children with DS.

CONCLUSIONS

All three studies provided statistically significant evidence indicating that activities using virtual reality can improve motor function in children with Down Syndrome. While the long-term effects remain to be unknown due to the lack of long-term follow up in these studies, the three studies yielded positive results for the VR-based activities as an effective form of therapy when compared to control groups or SOT. Since the aforementioned studies established the general conclusion that VR activities can improve motor function, further studies should be performed to determine the compared efficacy to the standard therapies today, and whether they should be considered as another form of standard therapy or recommended as an adjunct to the established standard therapies. It is important for future studies to keep track of outside activities during the duration of the intervention programs as there was a lack of control over outside activities in each of the studies, which could impact the results.

There remains to be a limited number of studies done specifically on children with DS; although, there are similar ongoing studies that have yet to be published. In a technologically advanced society, future studies should be open to more virtual reality-based technologies besides the Wii as they become available. Therapies that are enjoyable for children with DS should be kept a priority to encourage an active lifestyle guiding them to improved motor function and a better quality of life.

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