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Alexandra Perrot, Pauline Maillot, Agnès Le Foulon, Anne-Sophie Rebillat

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Title : Effect of exergaming on physical fitness, functional mobility and cognitive functioning in adults with Down Syndrome.

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Abstract

This study examined whether exergames could improve physical, functional, and cognitive functions in people with Down Syndrome. Twelve adults with DS, aged over 35 ($M = 50.35$, $SD = 7.45$), were randomly assigned to a Wii-based program ($n = 6$) or a control group ($n = 6$), and completed physical (Chair Stand Test, 6-Minute Walk Test), functional (TUG, TUDS), and cognitive tests (Corsi, Barrage tests). The experimental group completed a 12-week Wii-based program. There was high intervention adherence and, compared with the control group, greater improvements were observed in the Wii-based exercise intervention group in physical fitness and functional outcomes ($p < .05$), with no changes in cognitive outcomes.

Key words: Down Syndrome, exergames, physical activity, healthful aging

Introduction

Down Syndrome (DS) is a genetic disorder caused by the presence of all or part of a third copy of chromosome 21, typically associated with a range of impairments in intellectual and physical function (de Graaf et al., 2011). With progress in medicine and healthcare, and in particular better follow-up, the life expectancy of people with DS has increased from the age of 9 in 1929 to the age of 65 today (e.g., Bittles & Glasson, 2004; Fromage & Anglade, 2002). The DS population is subject to premature or accelerated aging (from the age of 35-40) (Zigman, 2013), which means that they experience certain conditions and physical features, common to typically aging adults, at an earlier age than the general population. As a result, the known deleterious effects of aging, on both physical and cognitive function, compound the syndrome impairments linked to DS itself, and have the potential to lead to activity limitations and participation restrictions for individuals with DS (Barnhart & Connolly, 2007).

The importance of encouraging physical activity in people with DS cannot be underestimated (Silva et al., 2017). Several studies show that regular physical activity allows people with DS to improve their cardiovascular fitness (Andriolo, El Dib, Ramos, Atallah & da Silva, 2010 ; Dodd & Shields, 2005), muscle strength and balance (Li et al., 2013) as well as daily living tasks, such as walking, rising from a chair and ascending/descending steps (Cowley et al., 2010). Some research also notes the benefits of physical activity on cognitive functions in young adults with DS (Chen, Ringenbach, Crews, Kulinna & Amazeen, 2015; Holzapfel et al., 2015; Ringenbach et al., 2008, 2016). Physical activity is highly recommended for aging people with DS. However, too few recommendations currently support this argument (INSERM report, 2016) and regular intentional physical activity still remains very low in the DS population.

Exergames (the combination of physical exercise and video games, e.g., Wii, Kinect Xbox), may be an appealing alternative to promote physical activity in DS (Silva et al., 2017).

Exergames are comprised of full-body physical exercises performed through active video games which require gross motor and visual-spatial coordination, balance, and energy expenditure, comparable to moderate-intensity physical activity (Mura, Carta, Sancassiani, Machado & Prosperini, 2017; Read & Shortell, 2011). Exergames are affordable and accessible (Wuang, Chiang, Su, & Wang, 2011), enjoyable and motivating (Mura et al., 2017), with high-levels of adherence (Maillot, Perrot, Hartley, 2012a, b; Tatla et al., 2014).

They allow participants to practice at home and to achieve the recommended aerobic exercise intensity and energy expenditure (Lanningham-Foster et al., 2009; LeGear et al., 2016).

Several studies have used exergames as physical activity stimulation for different clinical populations. In the field of aging, Maillot et al. (2012b; 2014) demonstrated cognitive and functional benefits for healthy aging samples after 24 x 1-hour training sessions on the Nintendo Wii. In the field of neurological disabilities, a systematic review and meta-analysis (Mura et al., 2017) documented that exergames were a highly-flexible tool in the rehabilitation of both cognitive and motor functions in adult samples suffering from various neurological disabilities and developmental neurological disorders such as multiple sclerosis, post-stroke hemiparesis, Parkinson's disease, dementia, dyslexia, as well as DS. Stanmore and collaborators suggest that exergames increase the ability of people with neurological disabilities to allocate cognitive resources in time and space by enhancing executive functions and visuo-spatial perception. The intrinsic characteristics of exergames, such as the highly variable environment, may well be partly responsible for cognitive improvements, by promoting learning processes dependent on executive function (Bavelier et al., 2012). In children with DS, Rahman (2012) showed that a 6-week Wii-Fit program could improve balance. Wuang, et al. (2011) demonstrated Wii benefits in improving motor proficiency and

sensorimotor functions. Berg et al. (2012) noted improvements in motor skills and postural control of one child with DS, following an 8-week Wii program of 4 x 20-minute sessions per week. In adults with DS, the one study to date, (Silva et al., 2017) suggests that Wii-based exercise can be an effective tool in improving physical fitness, functional mobility and motor proficiency.

The purpose of this study was to replicate and extend the research on exergame effects in adults with DS (Silva et al., 2017). The specific aims were to test the effects of a Wii-based exercise program on physical fitness outcome measures, functional mobility indicators and cognitive function (cognitive outcomes were not evaluated in the Silva study). We hypothesized that exergames would improve functional mobility and physical fitness in a DS sample. We also explored cognitive benefits given the results of several studies demonstrating the benefits of video games on cognition (e.g., Basak, Boot, Voss, & Kramer, 2008; Green & Bavelier, 2003), including individuals with intellectual disabilities (for review, Rodriguez Jimenez et al., 2015; Stanmore, Stubbs, Vancampfort, de Bruin & Firth, 2017).

Material and methods

Participants

Adults diagnosed with DS were recruited from a leading medical center specializing in DS, the Lejeune Institute in France. Inclusion criteria included authorization, by physicians of the institute, for the participants to engage in physical activity, and to be over the age of 35.

Participants were not included if they had a neuromusculoskeletal disorder, non-autonomous locomotion or were unable to understand the instructions. The institute geriatrician determined the individuals meeting these criteria, then proposed the study to them during a consultation. Twelve participants, between the ages of 35 and 64, were randomly allocated to either the experimental group (Wii-based exercise program; n = 6; 3 men; mean age = 49.3;

SD = 8.2) or the control group (no lifestyle changes; n = 6; 3 men; mean age = 51.4; SD = 6.7) and completed all of the intervention sessions and assessment procedures (Table 1). The study was approved by the local ethics committee of the Lejeune Institute.

Apparatus

For the exergame training we used the Nintendo Wii, a video game console with integrated motion-sensitive technology. The Wii Remote and the Nunchuk, the primary wireless console controllers, use a combination of built-in accelerometers and infrared detection to sense their positions in three-dimensional space when pointed at the LEDs in the Sensor Bar of the console. This design allows game players to control their avatars (graphical representations of the users) by using physical gestures as well as the traditional button presses. We also used the Nintendo Wii Balance Board, similar in shape to a body-weight scale with a flat rectangular platform (51.1cm x 31.6cm x 5.3cm). The player's weight and center of pressure trajectory are recorded by four force sensors during the game. It communicates wirelessly with the Nintendo Wii video game console. The video game display was an LCL projector (Panasonic Model PT-AX200E) and a portable screen (Epson ELPSC06), 102cm x 76cm. The heart rate (HR) was monitored with the aid of a standard chest lead connected to a digital pulse transmitter (SUUNTO T6 - software SUUNTO Training Manager, version 2.3.0.15).

Measures

All of the participants completed several tests in order to assess their functional mobility, muscular endurance, physical fitness and cognitive functions.

Timed Up and Go test and Timed Up and Down Stairs test

The Timed Up and Go test (TUG) and Timed Up and Down Stairs test (TUDS) were used to measure functional mobility. The TUG test (Podsiadlo & Richardson, 1991) uses the time that

a person takes to rise from a chair, walk three meters, turn around, walk back to the chair, and sit back down. For the TUDS test (Zaino, Marchese & Westcott, 2004), the participants were instructed to stand at the bottom of a flight of stairs, and to quickly, but safely, climb the stairs, turn around on the top step and descend the stairs until both feet landed on the bottom step.

30-second chair stand test

The 30-second chair stand test (Jones & Rikli, 2002) was used to measure muscular endurance. The participants were instructed to sit in the middle of a seat, feet flat on the floor and shoulder width apart, and arms crossed at the wrists and held close to the chest. From the sitting position, the subjects stand up completely and then sit back down completely. This is repeated for 30 seconds.

6-minute walk test

Physical fitness was assessed with the 6-minute walk test, which involves measuring the distance walked over a span of 6 minutes (American Thoracic Society, ATS, 2000; Lipkin, Scriven, Crake, & Poole-Wilson, 1986).

Heart rate

Heart rate (HR) was monitored at the outset of the study and during every Wii-based session. The purpose was to quantify the intensity of the physical exercise throughout the course of the program through the calculation of the proportion of the heart rate reserve according to the Karvonen method (1957). The heart rate reserve (HRR) is the difference between the estimated maximum heart rate ($MHR = 205 - 0.685 \times \text{Age}$) and the resting heart rate (RHR). The exercise program targeted moderate intensity, corresponding to 40-59% of the

estimated HRR (ACSM, 2006; Howley, 2001). The explanation of the results and calculations concerning the MHR, RHR and HRR can be found in Table 2.

Cognitive functions were assessed with a spatial span test and a selective attention test, from batteries of tests used to assess cognitive abilities in young children.

Corsi block tapping test

The spatial span test is the Corsi block tapping test (Wechsler, 1997) which assesses visuospatial short-term working memory. It involves mimicking a researcher as he / she taps a sequence of up to nine identical spatially separated blocks. The sequence starts out simply, usually using two blocks, but becomes more complex until the subject's performance suffers.

Stimulus barrage test

Selective attention was assessed with a stimulus barrage test (Korman, Kirk, & Kemp, 2012). The participants were asked to cross out the target stimuli (houses) among distractors (ladders, tables, planes), on an A4 sheet.

Procedure

The entire study lasted 14 weeks including both the pre- and post-test and program sessions. During the first session, the participants completed the tests. The functional, muscular and physical tests were performed by a master student in adapted physical activity under the supervision of the institute geriatrician. The cognitive tests were performed by the institute neuropsychologist. The participants in the experimental group then engaged in the Wii-based exercise program. The Wii-based exercise program consisted of 2 x 1-hour exergame sessions per week over a 12-week period, resulting in a total training time of 24 hours. Each session was individual and began with a warm-up and finished with a cool-down in order to reduce the risk of injury (e.g., Jones & Hammig, 2009). Each training session was divided into two

periods. During the first period, the participants played « Wii sports » (Wii Tennis and Wii Bowling). During the second period, the participants played « Wii Fit Plus » using the balance board to play Wii Soccer Headers, Wii Ski Jump, Wii Hula Hoop and the Wii Marbles games). The games were validated by the institute geriatrician as compatible with the characteristics of people suffering from DS, such as hypertonia, hyperlaxity and heart problems. Each training session was supervised by the master student in adapted physical activity. The participants were urged to try to increase the challenge level and improve their performance in each activity over the course of the training (Maillot et al., 2012). The participants in the control group committed themselves to maintaining their regular lifestyles and were not permitted to begin playing video exergame games or engage in any other physical activity over the 14 weeks of the study.

Statistical analysis

Given the small number of subjects, nonparametric statistical tests were conducted, with the use of Mann-Whitney tests. The first step in the analysis was to verify that the Wii-based exercise and the control group were equivalent at the outset of the program. Mann-Whitney tests were used to compare the pre-training scores on each measure. The central step in the analysis was to determine whether the training regimen resulted in greater change in the exergame group than in the untrained control group. We calculated the change score by subtracting the pre-test score from the post-test score for each measure and carried out the Mann-Whitney test on each component measure.

Results

None of the seven comparisons between the pre-training scores of the Wii-based exercise and the control group were statistically significant. The descriptive and inferential statistics are in Table 1. In the Wii-based exercise group, the number of completed sessions for the 6

participants ranged from 22 to 24; $M = 23.17$; $\max = 24$ sessions. The overall adherence was 96.5 %; 139 out of a possible 144 sessions completed by the participants (6 participants for 12 weeks / 2 sessions per week). The mean heart rate during the program indicated that none of the participants in the Wii-based group engaged in physical activity with moderate intensity (effort intensity below 40%, $M = 23.65$ %, $SD = 6.97$, between 14 and 34.8%; Table 2). Following the program, there were no statistically significant differences in the measured cognitive functions in the Wii-based exercise group compared with the control group ($p = n.s$). Improvement in the Wii-based exercise group was observed with large effect sizes for both functional measures ($p < .01$, *Cohen's d* = 2.23), muscular endurance ($p < .05$; *Cohen's d* = 1.74), and physical fitness ($p < .05$, *Cohen's d* = 1.39). The TUG and TUDS scores increased respectively by 15% and 12%, muscular endurance by 24%, and physical fitness by 5%. The descriptive and inferential statistics for the change scores are shown in Table 3.

Discussion

The purpose of the present study was to continue investigating the potential of Wii-based exercises as a mode of activity which could have physical, functional and cognitive benefits for adults living with DS. In this study sample, the Wii-based exercise program had a high level of adherence (96.5 %) consistent with prior studies reporting on individuals living with various neurological disabilities (Choi et al., 2014; Nilsagard, Forsberg & von Koch, 2013; Hugues, Flatt, Fu, Butters, Chang & Ganguli, 2014, Wuang, et al., 2011). The high adherence rate underscores the potential of exergames in terms of accessibility and intrinsic attractiveness for adults with DS. Interactive physical activity video games could be a functional alternative to physical activity alone (e.g., Hughes et al., 2009; Schutzer & Graves, 2004).

The first study hypothesis was that Wii-based exercise training would have functional and physical benefits. The results demonstrated significant benefits of exergame training in

functional, muscular endurance and physical fitness before and after training. The characteristics of the Wii games may help explain, in part, the effects (Maillot et al., 2012b; 2014). Games on the Wii Fit Plus, with the balance board, place great emphasis on balance skills, as well as muscular endurance in the lower limbs. "Wii sports" games, such as tennis, have also been able to reinforce physical fitness. It was encouraging to observe transfer benefits to physical and functional skills directly involved in functional abilities needed by older adults in everyday life, such as walking, getting up from a chair, and climbing stairs (e.g., Tomaszewski Farias et al., 2009). The beneficial effect of training on functional mobility (TUG, TUDS) and muscular endurance (30-second Chair Stand Test) is in line with the results of Stanmore et al (2017), demonstrating the exergame impact on motor functions such as balance performance and walking ability in people with neurological disabilities. Our results are also consistent with studies carried out with children (Berg et al, 2012; Wuang et al, 2011) and adults with DS (Silva et al., 2017), highlighting the exergame impact on functional mobility and strength. Meta-analytic findings on physical exercise intervention on a person's fitness also suggest that exercise interventions improve strength and balance in individuals with DS (Li, Chen, Meng How, & Zhang, 2013). Thus, exergames seem to be a relevant alternative to improve balance and muscular endurance in the lower extremities. This result is all the more important as muscle weakness (Carmelli et al., 2002), and postural control deficits (Shields et al., 2008) are characteristics of people affected by DS.

The improvements observed in physical fitness, assessed with 6 MWT, are in line with the work of Silva et al (2017), the first study which found a significant effect of exergame intervention on aerobic endurance in people with DS. These results also confirm existing literature on cardiovascular fitness, suggesting that cardiovascular exercise programs are beneficial and safe for people with DS (Dodd & Shields, 2005). This finding is important

since aerobic capacity is highly associated with cardiovascular disease and all-cause mortality (Sui et al., 2007; Desprès, 2016).

The second hypothesis of this study was that Wii-based exercise training could provide cognitive benefits to adults with DS. The results before and after training demonstrated no significant benefits of exergame training in selective attention and spatial span tasks. Previous studies have shown that exergames encompass most of the principles underlying learning processes such as high intensity repetition of task-oriented exercises, incrementally increased task difficulty, real-time feedback, motivation and rewarding transfer effects (Green & Bavelier, 2008; Kleim & Jones, 2008; Mura et al., 2017). The lack of cognitive improvement in our sample of adults with DS suggests that the cognitive benefits associated with exergaming are not fully understood and that there are multiple variables to be considered (timing, intensity, 'doseage', as well as different elements related to video game parameters). Previous studies report that video games have been successfully used with people with intellectual disabilities to improve several cognitive abilities (for review, Rodriguez Jimenez et al., 2015). The differences between the results could be due to the game profile or the cognitive function assessment. The games in the Rodriguez Jimenez et al review were mainly based on computer-assisted programs and stand-alone software, conducted in a laboratory created for therapeutic purposes, and targeting a particular function (e.g., writing skills, mathematics, working memory, word spelling). As a consequence, this concerns near transfer, as gains were obtained in specifically trained abilities. Our approach perhaps focused more on the far transfer notion. Additionally, there may have been measurement issues and that the neuropsychological tests we used were unable to detect subtle cognitive benefit due to a lack of sensitivity.

Our result is also inconsistent with previous studies on exergames, which emphasize a beneficial impact of exergame interventions on executive functions, attentional processing and visuospatial skills in adults with neurocognitive impairments (Stanmore et al., 2017). In this case, the differences in the results could be due to the participant profile. Stanmore and collaborators include cognitive impairment such as Parkinson's disease, mild cognitive impairment, sub-acute stroke, and schizophrenia. The cognitive impairment origin is therefore quite different from that of individuals with DS. But, again, there are multiple parameters that remain to be more fully investigated in relation to cognitive function and exergame effects.

The cardiovascular hypothesis, based on the improvement in oxygen transport with aerobic fitness, must also be addressed to explain the results concerning cognition. The increased oxygen availability for cerebral metabolic activity may be responsible for the improved neurocognitive performance which occurs with fitness training (e.g., Audiffren, André & Albinet, 2011) if the intensity is at least 40% of the estimated HRR (i.e., moderate; ACSM, 2006, Howley, 2001). Our results showed significant effects of training on the physical status, and more particularly on cardio-respiratory fitness (e.g., 6MWT). However, the HRR response did not fall within the ACSM recommended "moderate" intensity range. The effort intensity reported in this study was calculated in relation to the heart rate continuously evaluated during the training sessions. Since the sessions were composed of a variety of different exercises, alternation between peak heart rate time and rest time can be observed. Thus, the raised heart rate may have been underestimated. This result is not consistent with previous work which noted that exergames represent moderate activity (e.g., Graves et al., 2010; Maillot et al, 2012, 2014, Read & Shortell, 2011) and emphasizes that the program should be adapted in order to increase the intensity. It seems necessary to encourage more physical investment, either through incentives from the program manager or through the form

of play. Dance exergames also could be an interesting option (Brami, Trivalle, & Maillot, 2018).

There are a number of other study specific limitations to note. The sample was small and therefore it was likely underpowered weakening our ability to detect differences in cognitive functions. As a product, exergames were created to use therapeutically with individuals with DS. There is consequently a need for the development of specific and tailored programs for DS to establish accurately the type, intensity and frequency of games suitable and effective, with particular emphasis on the benefits to cognitive functions. A further limitation is that it is impossible to distinguish between the physical activity component and the video game component of the Wii-based exercise regimen as the possible source of improvement. We do not know if the effects observed are specific to exergames or simply the consequence of physical stimulation. A control group engaged in a standard physical activity program would be important for future investigations.

Conclusion

In conclusion, our study suggests that a Wii-based exercise program, combining cognitive and physical demands in the form of an intrinsically attractive activity, might well be considered as an effective way of promoting physical and functional improvements in adults with DS. Indeed, one current health issue in people with intellectual disabilities is the need to improve physical activity levels to offset the harm of sedentary lifestyles (Hilgenkamp, Reis, van Wijck, & Evenhuis, 2012; for review, Lotan, 2007), which increase with age (Finlayson et al., 2009). Thus, exergames could be an introduction to physical exercise and a complement to conventional physical activity for sedentary adults with DS. As emphasized by Maillot et al (2012, 2014), exergames possess many attributes which make them promising for widespread adoption. First, exergame play can be home-based, mitigating against environmental barriers

which could dissuade exercising. Second, the games allow choice among activities, which could lead to sustained play and foster autonomy. Finally, as noted by Mura et al in their meta-analysis (2017), exergames are safe, flexible and characterized by high adherence rates among people with neurological disabilities. The console is intuitive to use and accessible to people with intellectual disabilities. As Brown et al. (2011) suggested, it is better to use popular technology rather than design expensive assistive technology which creates false distinctions between people with typical development and those with intellectual disabilities. Some exergames have the advantage of simulating real-life situations and the possibility of repeating the same action as many times as necessary (Rodriguez Jimenez et al., 2015), with immediate feedback (Sastre, 1998) and a gradual increase in the level of challenge (Rodriguez Jimenez et al., 2015). Moreover, video game practice induces enhanced independence, increased self-determination, and greater self-esteem (Davies, Stock, & Wehmeyer, 2004). For all of these reasons, further research into exergaming and its positive effects for individuals with DS is warranted.

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