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Original Article

Review on the effects of physical activity on body composition and shape in people with Down Syndrome

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Abstract
People with Down Syndrome (DS) are often characterized by overweight or obesity and result to be less active than their peers. Both overweight/obesity and physical inactivity are considered important risk factors for health, thus augmenting mortality risk. World Health Organization recommends different training programmes for health benefits in different age ranges of general population. Effects of physical activity may be even more important in preventing health consequences in people with DS. In this context, we conducted a literature search of original articles, published between May 2010 and May 2020, on the effects of physical activity on body shape, body composition and bone quality in people with DS. We found twelve articles from all the continents, investigating the effects of different types of physical activity on body fat and lean masses, bone composition and anthropometry. In sum, the included studies reported improvements in body composition (with augmented lean mass and reduced fat mass), body size (with lower weight, waist circumference and BMI) and bone quality (in terms of bone mineral content and/or bone mineral density). In particular, body composition improves after conditioning and plyometric jumps training program, 2-wheel bicycle training, whole body vibration and swimming. Body shape is significantly improved by bicycle training, interval training and swimming. Finally, bone quality shows positive results after conditioning and plyometric jumps training program, whole body vibration and weight bearing exercise. Although different types of physical activity are investigated in people with DS, however the best training programme for this population is not clear. So future studies may focus on the comparison or combination of different types of physical activity to assess which one could maximize benefits.
Keywords: Down Syndrome; physical activity; body composition; body shape; bone quality.

Introduction

Down Syndrome (DS) is a disorder based on the presence of three copies of chromosome 21. Its prevalence varies from 1 on 400 to 1 on 1500 people in different countries [Kazemi et, 2016]. This neurodevelopmental disorder affects both mental and physical health. Indeed, it can be associated to different comorbidities [Bittles et al, 2007]. In particular, people with DS often have overweight or obesity [Bertapelli et al, 2016] and result to be less active than their peers [Shields et al, 2018]. Both overweight/obesity and physical inactivity are considered important risk factors for health, augmenting mortality risk [WHO, 2010]. World Health Organization (WHO) defines physical activity (PA) as “any bodily movement produced by skeletal muscles that requires energy expenditure” and recommends different training programmes for promoting health in different age ranges of general population [WHO, 2010]. Effects of PA may be even more important in preventing health consequences in people with DS, knowing that children with DS do not meet the PA guidelines for their age groups [Fox et al, 2019]. PA may have some effects on body shape (BS), body composition (BC) and bone quality (BQ). BS refers to people anthropometric characteristics, such as height (H), weight (W), waist circumference (WC), BMI and weight/height ratio (W/H). BC is assessed through instrumentations that allow to achieve total and regional fat mass (FM) and lean mass (LM), body water percentages and their distribution. BQ is expressed in quantitative terms as Bone Mineral Content (BMC) and Bone Mineral Density (BMD) or qualitative/structural terms.

A previous systematic review [Li et al, 2013] analysed the impact of physical exercise on fitness in people with DS. Authors found moderate to high improvements on muscle strength and balance. Shields and colleagues [Shields et al, 2018] reviewed the effect of regular exercise on oxidative stress in people with DS, concluding that there is uncertainty about that. Sugimoto and colleagues [Sugimoto et al, 2016] made a literature review on the effect of neuromuscular training on strength and mobility in people with DS, finding large to moderate effects on general strength, moderate to small effects on maximal strength, and small effect on functional mobility. No strong literature evidence is available about the effects of different types of PA on BS,

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BC and BQ. Saquetto and colleagues [Saquetto et al., 2018] showed in their systematic review that whole body vibration training achieved positive results on BC and BQ in children and adolescents with DS.
In this context, starting from a systematic literature review focusing on the original articles published in the last ten years, the present study aims to investigate: (i) the main effects of PA on BS, BC and BQ; (ii) the comparison between results from different type of exercises.

Methods

We searched two online databases: PubMed (PM) and Web of Science (WoS). The selection of articles was made through ("Down Syndrome") AND "Exercise"[Mesh] for PM database and through ("Down Syndrome") AND (("Physical Activity") OR ("Physical Exercise")) for WOS. Moreover, we checked the reference list of all the screened long paper.

Papers Selection Criteria

The analysis of databases was made through the following criteria: (i) articles published between May 2010 and May 2020, in order to overview the most recent literature evidence; (ii) original articles, excluding reviews, commentaries, posters and proceeding papers; (iii) only full paper English written articles. After the first screening, two authors reviewed independently the founded articles with their title and abstract, in order to check the matching with the research aim. They selected papers aiming at investigating the effect of PA on BS, BC and BQ, and combined the articles, excluding duplicates. Then, they checked the long paper of every of these articles excluding: (i) articles dealing with people with DS and other severe comorbidity (potentially influencing results); (ii) articles assessing the effects of PA on other outcomes; (iii) articles considering animal models for DS.

Data extraction

From the selected papers the following data were extracted: (i) year of publication; (ii) participant characteristics (number, nationality, age, sex); (iii) assessed outcome(s); (iv) control; (v) type of PA; (vi) duration and frequency; (vii) results.

Results and discussion

The review process is shown in the flow diagram in Figure 1, using the PRISMA guidelines [10].

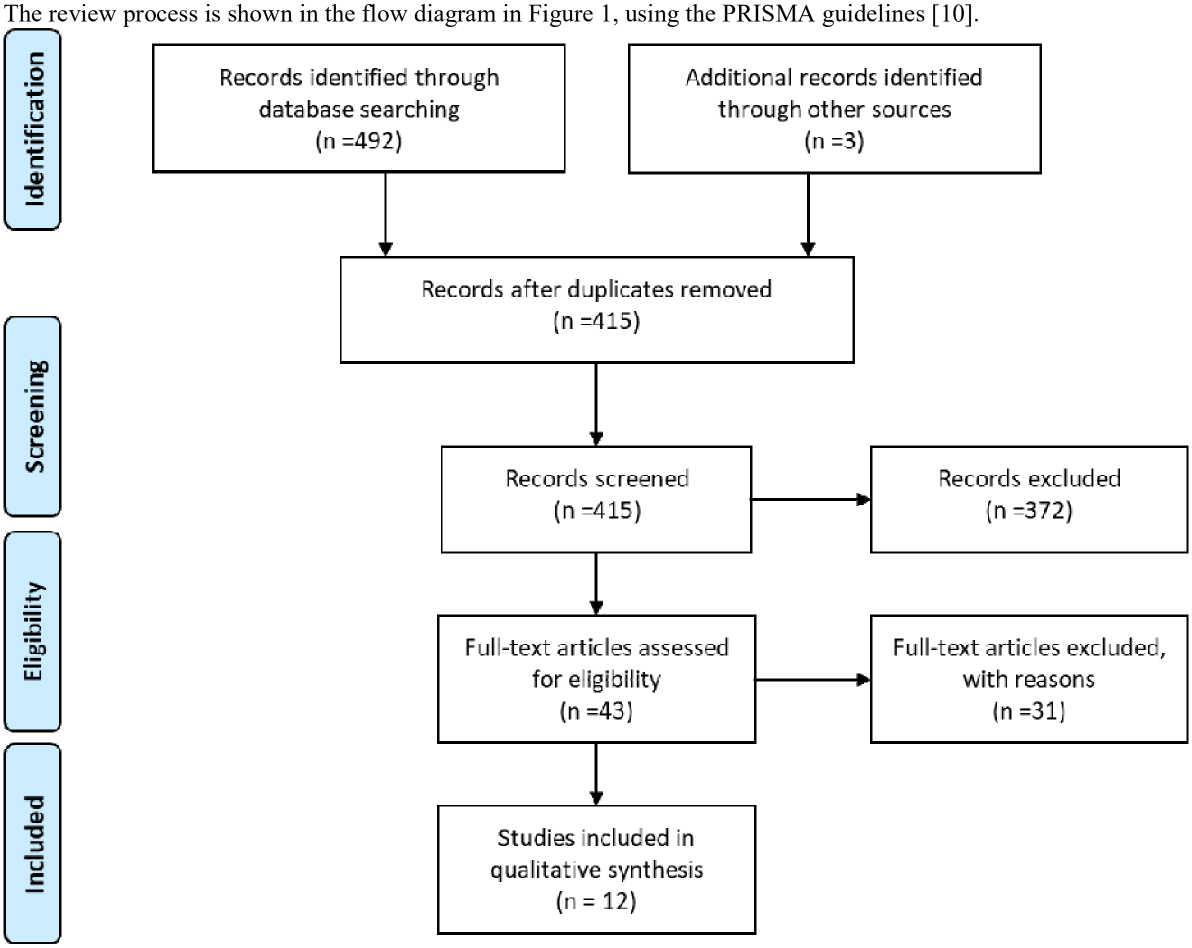

Figure 1: Flow diagram representing the literature review process

After applying the paper selection criteria mentioned before, we checked 43 long papers and excluded: 2 articles dealing with people with DS and other severe comorbidity (influencing results); 27 articles assessing the effects of PA on other outcomes; 2 articles considering animal models for DS. Finally, the selected articles were 12. Table 1 shows their main characteristics.

Table 1: selected articles main characteristics (n.a.: not available)

Author and year	Nation of participants	Total Participants Number (number of M: males)	Age range [years]	Participants in Experimental Group (EG)/Control Group (CG)
González-Agüero et al., 2011	Spain	26 (15 M)	10-19	13/13
Ulrich et al., 2011	United States of America	46 (20 M)	8-15	19/27
González-Agüero et al., 2012	Spain	28 (15 M)	10-19	14/14
González-Agüero et al., 2013	Spain	30 (19 M)	12-18	16/14
Reza et al., 2013	Iran	48 (n. a.)	7-12	n. a.
Matute-Llorente et al., 2015a	Spain	25 (17 M)	12–18	11/14
Matute-Llorente et al., 2015b	Spain	26 (15 M)	12–18	13/13 (non DS)
Shields et al., 2015	Australia	14 (8 M)	7-17	Observational (no CG)
Boer & Moss, 2016	South Africa	42 (25 M)	18-50	26 (13 CAT, 13 IT)/16
Pitchford et al., 2018	United States of America	39 (21 M)	12-18	22/17 (non DS)
El et al., 2019	Turkey	26 (15 M)	5-17	Cohort (no CG)
Suarez-Villadat et al., 2020	Spain	45 (25 M)	12-15	15/30

As we can see from Table 1, available studies cover all the continents. Most of the studies were randomized control trial with DS people in the control group, apart from [Matute-Llorente et al, 2016] that used typically developed peers as control. Moreover, there was a cohort study [El et al, 2015] and two observational studies [Shields et al, 2017; Pitchford et al, 2018]. The number of participants for each study ranged from 14 to 48 (with a mean value of 32.9), with a 56.2% of prevalence of male sex. Almost all the included studies focused on children and adolescents with DS, only one study [Boer & Moss, 2016] focused on adults. Table 2 shows the main detailed information of the selected studies.

Table 2: detailed information of the selected studies, with outcomes, type, duration and frequency of PA and main results, only statistically significant results are reported (BC: Body Composition, BIA: Bioelectrical Impedance Analysis, BMC: Bone Mineral Content, BMD: Bone Mineral Density, CG: Control Group, CVf: cardiovascular fitness, DXA: Dual-energy X-ray Absorptiometry, EG: experimental Group, FM: Fat Mass, H: height, LM: Lean Mass, PA: physical activity, qCT: quantitative computed tomography, Sk: Skinfold, W: weight, (+): statistically significant improvement)

Author and year	Outcomes (instrumentation)	Type of PA	Duration and frequency	Main results
González-Agüero et al., 2011	BC (DXA)	Conditioning and plyometric jumps training program	21 weeks (25 min, 2/week)	EG: (+) total and lower limbs LM CG: (+) upper limbs FM
Ulrich et al., 2011	H, W, FM (Sk caliper)	2-wheel bicycle	5 days	EG: (+) BMI and FM (after 12 months of follow-up)
González-Agüero et al., 2012	Total and regional BMC and total LM (DXA)	Conditioning and plyometric jumps training program	21 weeks (25 min, 2/week)	EG: (+) total and hip BMC, total LM
González-Agüero et al., 2013	BC (DXA)	Whole body vibration	20 weeks (15-20 min, 3/week)	EG: (+) upper limbs FM
Reza et al., 2013	BMD right proximal femoral head (DXA)	Weight bearing exercise	4 months (45 min, 3/week)	EG: (+) BMD
Matute-Llorente et al., 2015a	BMC and BMD (DXA) and bone structure	Whole body vibration	20 weeks (20 min, 3/week)	EG: (+) total and regional BMC and BMD

	variables (qCT)			
Matute-Llorente et al., 2015b	BMC and BMD (DXA)	Whole body vibration	20 weeks (20 min, 3/week)	EG: (+) subtotal and regional BMC and BMD
Shields et al., 2015	BMI, WC	PA (accelerometer)	8 days consecutive monitoring	PA: no correlation BMI and WC CVf: significant correlation BMI and WC
Boer & Moss, 2016 [19]	W, H, WC, BMI, BC (BIA)	Continuous aerobic training (CAT) vs. interval training (IT)	12 weeks (2-3/week)	EG: IT: (+) W and BMI
Pitchford et al., 2018	BMI, BC (DXA)	PA (accelerometer)	7 days consecutive monitoring	moderate-to-vigorous PA: Moderate association FM%
El et al., 2019	BC (BIA)	PA (Eurofit battery)	-	PA: Negative correlation FM and BMI.
Suarez-Villadat et al., 2020	BMI, WC, W/H, FM (triceps, subscapular, suprailiac and thigh Sk)	Swimming	36 weeks (3/week)	(+): BMI, WC, W/H, triceps and suprailiac Sk

Table 2 underlines that, about BC, three studies used Dual-energy X-ray Absorptiometry (DXA) [González-Agüero et al, 2011; González-Agüero et al, 2012; González-Agüero et al, 2013], two Bioelectrical Impedance Analysis (BIA) [Boer & Moss, 2016; El et al, 2015] and two studies assessed skinfolds (Sk) only to evaluate fat mass (FM) [Ulrich et al, 2011; Suarez-Villadat et al, 2020]. Five studies [Ulrich et al, 2011; Shields et al, 2017; Boer & Moss, 2016; Pitchford et al, 2018; Suarez-Villadat et al, 2020] assessed BS in terms of H, W, WC, BMI and/or W/H, using hand-held measurements.

No study assessed BS through automatic and user-friendly methods, such as photogrammetry [Grazioso et al, 2019a; 2019b], allowing an instantaneous 3D body measure, that could help in having accurate BS measurements with high patient compliance (important in people with disabilities, such as DS). Moreover, four studies used DXA to evaluate the effect on BQ (BMC: Bone Mineral Content and BMD: Bone Mineral Density) [González-Agüero et al, 2012; 2013; Reza et al, 2013; Matute-Llorente et al, 2015; 2016], with [Matute-Llorente et al, 2015] even using quantitative computed tomography (qCT).

PA varied in the interventive experimental studies, using both aerobic and strength training programmes. They includes from a conditioning and plyometric jumps training program [González-Agüero et al, 2011; 2012] to 2-wheel bicycle [Ulrich et al, 2011], from whole body vibration (WBV) [Matute-Llorente et al, 2015; 2016; González-Agüero et al, 2013] to weight bearing exercise (WBE) [Reza et al, 2013], from swimming [Suarez-Villadat et al, 2020] to the comparison among continuous aerobic training (CAT) and interval training (IT). The duration of the training programme varied from 12 to 36 weeks for the experimental studies, apart from [Ulrich et al, 2011] that used a 5-day training period with a follow-up at 12 months. Moreover, the three observational studies assessed PA through Eurofit battery [El et al, 2015] or accelerometers [Shields et al, 2017, Pitchford et al, 2018].

Accelerometers, as in a previous review [Fox et al, 2019], are only used to quantify PA, but they could even achieve useful information about PA quality and characteristics. Indeed, specific indices are often used to monitor PA in typically developed people [Caporaso & Grazioso, 2020; Caporaso et al, 2020]. This could be an interesting field to improve objective evaluation of PA in DS people and to regularly monitor it to personalize the training program.

In the complex, BC improves after conditioning and plyometric jumps training program (with augmented lean mass, LM) [González-Agüero et al, 2011; 2012], 2-wheel bicycle training [Ulrich et al, 2011], WBV [González-Agüero et al, 2013] and swimming [Boer et al, 2018] (with reduced FM). BS is significantly improved in terms of BMI by bicycle training [Ulrich et al, 2011], IT [Boer & Moss, 2016] and swimming [Suarez-Villadat, et al, 2020], with additional effects on W [Boer & Moss, 2016] and WC and W/H [Suarez-Villadat, et al, 2020]. Finally, BQ shows positive results in terms of BMC and/or BMD after conditioning and plyometric jumps training program [González-Agüero et al, 2012], WBV [Matute-Llorente et al, 2015; 2016] and WBE [Matute-Llorente et al, 2015].

These results show stronger evidence of PA impact on BS and BC than the ones reported by Li et al. [Li et al, 2013], that showed no significant improvement or few results, basing only on three studies. A specific multifactorial programme, including both strength and aerobic components, could be tested to achieve improvements on all the considered outcomes.

Moreover, Boer and colleagues even published a follow-up study [Boer et al, 2018], after three months of detraining following the [Boer & Moss, 2016]. They found significant reductions in maximal aerobic capacity, time to exhaustion and functional tests, underlying the negative effects of detraining.
From the observational studies emerged that PA negatively correlates with FM and BMI [Pitchford et al, 2018, El, et al, 2015], while cardiovascular fitness significantly correlates with BMI and WC [Shields et al, 2017].

Conclusions

This study concerns the investigation of the effects of PA on BS, BC and BQ. The available results, based on twelve studies from all over the world, show that regular PA may have a strong positive impact on BC (with augmented LM and reduced FM), BS (with lower W, WC and BMI) and BQ (in terms of BMC and/or BMD) in people with DS. In particular, BC improves after conditioning and plyometric jumps training program, 2-wheel bicycle training, WBV and swimming. BS is significantly improved in terms of BMI by bicycle training, IT and swimming. Finally, BQ shows positive results in terms of BMC and/or BMD after conditioning and plyometric jumps training program, WBV and WBE. Although different types of PA are investigated in people with DS, however the best training programme for this population is not clear. So future studies may focus on the comparison or combination of different types of PA to assess which one could maximize benefits. Apart from that, we are confident that a regular exercise should be proposed to every person with DS to allow body improvements and health preventive strategies, to avoid the negative effect of inactivity such as obesity/overweight and their metabolic impact.

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