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## Review

# Health-related physical fitness in children and adolescents with Down syndrome and response to training

A. González-Agüero<sup>1,2</sup>, G. Vicente-Rodríguez<sup>1,2</sup>, L. A. Moreno<sup>1,3</sup>, M. Guerra-Balic<sup>5</sup>, I. Ara<sup>1,4</sup>, J. A. Casajús<sup>1,2</sup>

<sup>1</sup>GENUD (Growth, Exercise, NUtrition and Development) Research Group, University of Zaragoza, Zaragoza, Spain, <sup>2</sup>Faculty of Health and Sport Sciences, Huesca, University of Zaragoza, Zaragoza, Spain, <sup>3</sup>School of Health Science, University of Zaragoza, Zaragoza, Spain, <sup>4</sup>University of Castilla La Mancha, Toledo, Spain, <sup>5</sup>Fundacio Blanquerna, University Ramon Lull, Barcelona, Spain  
Corresponding author: José Antonio Casajús, GENUD Group; Ed. Cervantes. Corona de Aragón St. 42, 2nd floor, 50009 Zaragoza, Spain. E-mail: joseant@unizar.es

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**Physical fitness is related to health at all ages. Information about physical fitness in the Down syndrome (DS) population, however, is scarce, especially when we consider children and adolescents. A review of the current data available on this topic would be both timely and important as it would serve as a starting point to stimulate new research perspectives. The data we reviewed from the literature showed a general trend toward lower values of physical fitness parameters and worse body composition variables in children and adolescents with DS compared with the population without intellectual disability (ID) or even with the population with**

**ID without DS. Notably, children and adolescents with DS have been described as less active or overprotected; however, these factors may not be the cause of their poor physical fitness. Many of the training programs carried out in children and adolescents with DS did not yield the desired responses, and the reasons are still unknown. The purpose of this review is to summarize the current available literature on health-related physical fitness in children and adolescents with DS, and the effect of training on these variables. From the literature available, it is clear that more data on this population are necessary.**

Down syndrome (DS) is a condition that is accompanied with intellectual disability (ID) and associated with abnormalities in chromosome 21. Although the triplication of the chromosome is the most common defect, translocation and nondisjunction are also described (Pueschel, 1990). Estimation of DS is about one out of 700–1000 live births (Smith, 2001; Roizen & Patterson, 2003), and its life expectancy is increasing, from an average of 9 years of age in 1929 (Bittles & Glasson, 2004) to 55 years and older in the present day (Smith, 2001; Glasson et al., 2002).

More than 80 clinical characteristics have been described in individuals with DS, including congenital heart diseases, which is present in approximately 40% of individuals with DS (Pueschel, 1990). Pueschel and Werner (1994) found the mitral valve prolapse to be the cause of around 80% of abnormal echocardiographies in their sample of 36 home-reared young individuals with DS. However, the most common congenital heart disease is the atrio-ventricular septal defect, with a prevalence of 45%, followed by a ventricular septal defect in 35% and an isolated atrial septal defect in 8% of the cases (Frid et al., 1999; Freeman et al., 2008; Vis et al., 2009). Leukemia is another serious disease that occurs with

a higher frequency in children with DS than in their peers without DS, although individuals with DS have a decreased risk of developing solid tumors in all age groups (Hasle et al., 2000).

Evidence suggests that some of the clinical characteristics of DS (Pitetti et al., 1993) such as muscle hypotonicity, hypermobility of the joints or ligamentous laxity, light to moderate obesity, an underdeveloped respiratory and cardiovascular system and short stature (short legs and arms in relation to torso) are related to exercise. In addition, poor balance and perceptual difficulties have been also described (Winnick, 1995). Moreover, characteristics associated with hypotonia and hypermobility, for example lordosis, ptosis, dislocated hips, kyphosis, flat pronated feet, forwarded head and atlantoaxial instability have been observed in this population (Winnick, 1995; Pueschel, 1998). One of the most important concerns regarding sport participation is atlantoaxial instability, as participation in contact sport activities are contraindicated in those cases (Pueschel, 1998).

Owing to their clinical characteristics, both youths and adults with DS have lower levels of cardiovascular fitness compared with matched controls without DS (Fernhall et al., 1996, 2001; Guerra et al.,

2003a, b). Studies on DS children indicate a more sedentary lifestyle and more time spent indoors compared with their siblings without DS (Sharav & Bowman, 1992); however, Frey et al. (2008) attributed this to paternal overprotection. Low levels of physical fitness may induce functional deterioration due to an increase in the prevalence of overweight or obesity, as well as a reduction in bone mass development, which may ultimately result in the aggravation of their clinical manifestations (Fig. 1).

Physical activity (PA) and sport participation produce many health-related benefits in children and adolescents: PA improves cardiovascular fitness (Vicente-Rodriguez et al., 2005), it contributes to a healthier lifestyle (Stewart et al., 2003), and it may enhance the antioxidant defense system (Franzoni et al., 2005) which delays cell aging. In children, regular PA and sport, as well as physical fitness levels, are associated with increased and higher accumulation of bone mass (Vicente-Rodriguez, 2006), fat mass reduction (Ara et al., 2004, 2007) and a physiological and healthy adiposity development (Ara et al., 2006). PA interventions have also been shown to benefit children with leukemia (San Juan et al., 2007). Interestingly, the payback is not purely a physical one, as benefits in social factors associated with sport participation are also described (Andriolo et al., 2005). Therefore, taking into account all these separate studies, it is suggested that PA could be a potential factor in helping children with DS improve their quality of life.

According to the American College of Sports Medicine (ACSM), health-related physical fitness includes body composition, aerobic capacity, muscular strength and flexibility (Heyward, 2006), although flexibility is not a priority for the DS population due to fact that augmented flexibility is predominant in this group (Pitetti et al., 1993).

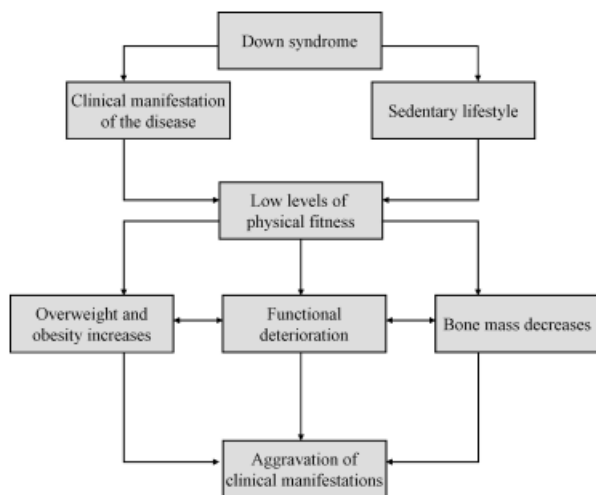


Fig. 1. Relationship between Down syndrome physical fitness and clinical manifestation.

In order to stimulate more research in this field, this paper aims to review the current literature on physical fitness, body composition and also the effects that training has on children and adolescents with DS.

## Method

### Inclusion criteria

The inclusion criteria for this review were as follows: (a) physical fitness or body composition had to be the main topic of each study but not necessarily PA; (b) the studies had to include participants with DS and not only with ID; (c) participants aged between 10 and 18 had to be at least 10% of the population studied; (d) only papers written entirely in English were considered.

### Data sources

Journal articles were sourced from MEDLINE (1965–present) and SPORT Discus (1975–present). The keywords used to identify the articles were “Down syndrome,” to restrict the population on this review; these terms were combined with “exercise,” “body composition,” “physical fitness” and “training” to identify the articles on the topic of this review. This produced a total of 101 citations from both databases.

### Exclusion

The inclusion criteria were applied to the 101 citations by two authors independently; in case of a disagreement, all authors reviewed until a consensus was achieved. Of the 101 citations, 22 journal articles fulfilled the inclusion criteria. The remaining 81 citations were excluded for the following reasons: 11 were duplicated, 57 did not include physical fitness or body composition as their main topic, five were not journal articles and six had participants outside our established population range.

### Data extraction

All the studies were evaluated independently by the authors of this review. General information about the title of the study, author(s), journal and publication details were extracted. Characteristics of the participants (age, sex), control group (if available), data source and results were also extracted. Studies including a training program were explained in detail.

## Health-related physical fitness in children and adolescents with DS

All the studies concerning health-related physical fitness in children and adolescents with DS included in this review are summarized in Table 1.

### Body composition

Body mass index (BMI) and different body compartments, such as body fat, lean mass and bone mass [bone mineral content (BMC) and bone mineral density (BMD)] have been studied in children and adolescents with DS. Children with DS were

Table 1. Studies concerning health-related physical fitness including children and adolescents with Down syndrome

Authors	Participants (age)	Control group	Data source	Results*
Guerra et al. (2009)	19; 7 F, 12 M (14.8 ± 3)	No	Wingate anaerobic test	Reliability between tests was questionable DS adolescents showed low levels of WANT performance compared with published data
Baynard et al. (2008)	133 (9–45)	180 subjects with MR without DS; 322 subjects without MR	Data collection of the last 20 years using the validated treadmill test	Lower relative and absolute ( $\dot{V}O_{2peak}$ ) across all age groups
Halaba et al. (2006)	7 (9.6 ± 1.8)	24 subjects without MR	Ultrasound at hand phalanges	( $\dot{V}O_{2peak}$ ) did not change after 16 years Lower amplitude-dependent speed of sound over ages and remained stable with time
Baptista et al. (2005)	67; 33 F, 34 M (14–40)	67 subjects without MR	DXA	Female group had lower muscle mass, higher percentage of body fat and BMI Lower BMC, BMD and volumetric BMD in the upper and lower limbs and the lumbar spine in the whole group with DS
Baynard et al. (2004)	13; 6 F, 7 M (18.5 ± 2.3)	17 subjects with MR without DS	Individualized treadmill test to exhaustion	Determination of VT is difficult in this population
Pitetti and Fernhall (2004)	119; 57 F, 62 M (14.8 ± 2.6)	395 subjects with MR without DS; 607 subjects without MR	20 m shuttle run test	Lower ( $\dot{V}O_{2peak}$ ), ( $\dot{V}E_{peak}$ ), $HR_{peak}$ and $RER_{peak}$
Fernhall et al. (2003)	89 (14.5); 47 obese	84 MR without DS; 22 obese	Treadmill test	Subjects with DS showed lower running performance than subjects without DS, with or without MR
Guerra et al. (2003a, b)	26, 11 F, 15 M (15.3 ± 2.7)	No	20 m shuttle run test and treadmill test	Lower maximal HR in the DS group, no differences between obese and nonobese
Fernhall et al. (2001)	97 (9–46)	179 subjects with MR without DS 196 subjects without MR	Multicenter study with the treadmill test	Controlled for maximal HR, no changes in aerobic capacity between obese and nonobese DS Regression formula for children and adolescents with MR is not valid in their sample of adolescents with DS Prediction formula for $HR_{max}$ Lower ( $\dot{V}O_{2peak}$ ), ( $\dot{V}E_{peak}$ ), $HR_{peak}$ and $RER_{peak}$

Table 1. (continued)

Authors	Participants (age)	Control group	Data source	Results*
Mercer and Lewis (2001)	17; 11 F, 6 M (11.2 ± 2.4)	17 subjects without MR	Anthropometric measurements Hand-held dynamometer to evaluate muscle forces	Higher BMI and percentage of body fat Lower mean peak torque values for hip abduction and knee extension Reliability high between test (0.89–0.95) Weight, height, gender, BMI and activity levels were significant predictors for peak torque production in DS Validation of the formula to predict ( $\dot{V}O_{2peak}$ ) with the 20 m shuttle run test Formula to predict ( $\dot{V}O_{2peak}$ ) Validation against 600-yard run-walk, 20 m shuttle run and a modified 16 m shuttle run tests
Fernhall et al. (2000)	17; 8 F, 9 M, all with MR (13.7)	No	Treadmill test and the 20 m shuttle run test	No differences in fat-free mass
Fernhall et al. (1998)	34; eight with DS (14.3 ± 2.3)	No	Treadmill test and field tests (600-yard run-walk, 20 m shuttle run and 16 m shuttle run)	No differences in BMI Less active, more time indoors Lower BMD at the lumbar spine Delay in the distribution curve of BMD against ages
Luke et al. (1996)	10; 6 F, 4 M (8.8 ± 2.5)	10 subjects without MR	Anthropometric measurements, bioelectrical impedance and deuterium dilution	Treadmill test validation High reliability between the two tests ( $r = 0.94$ ) Lower $\dot{V}O_{2max}$ Shorter performance time and lower maximal workload Blood pressure did not increase regularly
Sharav and Bowman (1992)	30; 16 F, 14 M (F5.1 ± 2.8, M4.1 ± 2.5)	Siblings without DS	Anthropometric measurements, accelerometers and questionnaires DPX	No differences in BMI
Kao et al. (1992)	10; 3 F, 7 M (10–16)	Reference without MR		
Fernhall et al. (1990)	14; 3 F, 11 M (17.7)	No	Treadmill tests	
Eberhard et al. (1989)	10; 3 F, 7 M (14.8 ± 2.2)	Eight subjects without MR	Bicycle ergometry test	

\*In the results, all the comparisons are as follows: the group with DS compared with the group without DS (with or without MR) when existing.

F, female; M, male; DS, Down syndrome; MR, mental retardation; SD, standard deviation; DXA, dual energy x-ray absorptiometry; DPX, dual photon x-ray absorptiometry; BMI, body mass index; BMC, bone mineral content; BMD, bone mineral density;  $\dot{V}O_2$ , oxygen consumption; VE, ventilation; HR, heart rate; RER, respiratory exchange ratio.

described as less active and more prone to spending more time indoors, but no differences were found in the BMI values between children with DS and their siblings without ID (Sharav & Bowman, 1992). However, several other investigations, some of them including adults in the sample, have shown a tendency toward a higher BMI and percentage of body fat in groups with DS compared with those without ID (Mercer & Lewis, 2001; Baptista et al., 2005). It is notable that Luke et al. (1996) found no difference in the fat-free mass (measured with deuterium dilution and other methods) between children with and without DS. On the other hand, Baptista et al. (2005), estimating the total muscle mass as suggested by Heymsfield et al. (1990) using data from Dual energy x-ray absorptiometry, found lower values in both males and females with DS compared with males and females without ID.

Common to both children and adolescents with DS is a lower BMC and BMD at the lumbar spine (Kao et al., 1992; Baptista et al., 2005) and the upper and lower limbs (Baptista et al., 2005). Furthermore, lower volumetric BMD has been found in other areas such as the upper and lower limbs (Baptista et al., 2005).

Similarly, a study with ultrasonography found lower amplitude-dependent speed of sound, which depends on BMD, in 24 children with genetic disorders (including seven children with DS) compared with age-matched children without ID; however, the difference remained stable with time (Halaba et al., 2006), indicating low bone mass but normal development.

In conclusion, although there have been several important reports on body composition in children and adolescents with DS, more studies are required to describe not only the body composition of children and adolescents with DS, but also the effect of exercise on the lean, fat and bone compartments in this population.

### Cardiovascular fitness

Lower levels of cardiovascular fitness have been reported several times in children and adolescents with DS compared with their peers without DS, with or without ID (Eberhard et al., 1989; Fernhall et al., 1996, 2001; Guerra et al., 2003a, b; Pitetti & Fernhall, 2004; Baynard et al., 2008).

Eberhard et al. (1989) found a lower maximal oxygen consumption ( $\dot{V}O_{2\max}$ ), a shorter time and a lower maximal workload in children with DS compared with the control, age-matched children. In the last two decades, considerable progress has been made in the assessment of cardiovascular fitness in children and adolescents with ID but specifically with DS. Fernhall et al. (1990) developed a validated

treadmill test method for adolescents and adults with ID (including DS). The findings of the study showed a high reliability coefficient of 0.94 between two repeated tests. In further studies, Fernhall et al. (1998) developed a regression equation to predict a peak oxygen consumption ( $\dot{V}O_{2\text{peak}}$ ) in children and adolescents with ID (including DS) with field tests (600-yard run-walk, 20 m shuttle run and a modified 16 m shuttle run), and in 2000, they validated the equation again with the 20 m shuttle run test (Fernhall et al., 2000). However, Guerra et al. (2003a, b) found that the regression equation to predict ( $\dot{V}O_{2\text{peak}}$ ) in children with ID was not valid for their sample of adolescents with DS, and attributed this to the low number of children with DS in the sample of Fernhall. Also, Fernhall et al. (2001) developed an equation to predict the maximum heart rate ( $HR_{\max}$ ) in individuals with ID (including children and adolescents with DS). Baynard et al. (2004) and Fernhall et al. (2001) found lower ( $\dot{V}O_{2\text{peak}}$ ), peak ventilation ( $\dot{V}E_{\text{peak}}$ ),  $HR_{\text{peak}}$  and peak respiratory exchange ratio ( $RER_{\text{peak}}$ ) in children and adolescents with DS compared with peers without DS, with or without ID. In more recent studies, Baynard et al. (2008) divided their sample of a multicenter study into age groups, and found lower relative and absolute ( $\dot{V}O_{2\text{peak}}$ ) across all age groups in the individuals with DS compared with both groups (the group with ID without DS and the group without ID). Crucially, for the DS group, their ( $\dot{V}O_{2\text{peak}}$ ) did not change after 16 years of age. Pitetti and Fernhall (2004) found lower running performance in their subjects with DS compared with the ones without DS, with or without ID.

Low cardiovascular fitness is considered to be a risk factor for cardiovascular diseases, and can result in a shortened lifespan for children and adolescents with DS. However, due to the lack of studies that include only the pediatric population in this regard, more studies are required to corroborate this assumption.

### Strength

Correct levels of muscular strength are related to health (Heyward, 2006) and help people to be more autonomous and independent; however, especially in old age, it is difficult to maintain those levels (Frontera et al., 1991; Brooks & Faulkner, 1994). As the lifespan of the DS population is increasing, it is important to study the actual level of strength in this population and, if necessary, promote programs to improve it.

Only one study related to muscular strength fulfilled all the inclusion criteria for this review. Mercer and Lewis (2001) found a lower mean peak torque for hip abduction and knee extension for children

Table 2. Studies concerning physical training including children and adolescents with Down syndrome

Study	Participants	Control	Data source	Training	Results*
<i>Aerobic training</i> Ordoñez et al. (2006)	22 M (16.2 ± 1)	No	Anthropometric measurements	12-week physical activity program; intensity level on the basis of HR; 30–60 min per session, three sessions per week	Significant reduction in the fat mass percentage
Varela et al. (2001)	16 M (21.4 ± 3)	Eight subjects with DS, no exercise	Anthropometric measurements, treadmill or rowing ergometer peak-graded exercise test	16-week rowing ergometer; intensities 55–70% ( $VO_{2peak}$ ); 15–25 min per session; three sessions per week	No differences in cardiovascular or physiological responses Exercise group achieved higher levels of work performance No changes in cardiovascular capacities Exercise group improved the time to exhaustion and grade
Millar et al. (1993)	14; 3 F, 11 M (17.7 ± 2.9)	Four subjects with DS, no exercise	Walking treadmill test	10-week walking jogging exercise program; 65–75% $HR_{max}$ ; 30 min per session; three sessions per week	Weight training had greater effects in all muscular strength tests than the strength treatment
<i>Strength training</i> Weber and French (1988)	14; 3 F, 11 M (13–18)	No	Ten exercises to evaluate muscular strength: Dorsi pull down, leg press, upright row, leg extension, shoulder press, calf raise, arm curl, leg curl, chest press and dead lift	Two groups: group A performed a 6-week (three times per week) weight training treatment at 80% 1 RM; group B performed a 6-week (three times per week) strength treatment 15 min per session	BMI did not change Decreased HR and RER in all the stages of the treadmill test Higher anaerobic power and strength in the trunk, the upper and lower limbs
<i>Combined aerobic and strength training</i> Lewis and Fragala-Pinkham (2005)	1 F (10.5)	No	Anthropometric measurements, submaximal treadmill stress test, modification of Margaria-Kalamen test, 10 RM to upper and lower limbs	6-week combined aerobic and strength training Aerobic intensity 60–80% $HR_{max}$ , 10–60 min per session; two to three sessions per week Strength intensity increased by the number of repetitions and weight; 10–45 min per session, two to three sessions per week	

\*In the results, all the comparisons are as follows: the group with DS compared with the control group when existing.

F, female; M, male; DS, Down syndrome; MR, mental retardation; SD, standard deviation; BMI, body mass index;  $VO_2$ , oxygen consumption; HR, heart rate; RER, respiratory exchange ratio; RM, maximum repetition.

and adolescents with DS compared with their peers without ID, where weight, height, gender, PA level and BMI serve as significant predictors for peak torque production in children and adolescents with DS.

### Effects of training in children and adolescents with DS

From the wealth of articles in the literature, it can be concluded that children and adolescents with DS have lower levels of strength and cardiovascular fitness, coupled with higher levels of body fat when compared with their peers without DS, both with and without ID.

As the PA level is a significant predictor of strength in the population with DS (Mercer & Lewis, 2001), testing whether supervised exercise interventions could improve muscular strength, cardiovascular fitness and body composition, which could also result in a concomitant health enhancement, is an important issue that needs to be addressed.

All the studies related to the effect of physical training programs in children and adolescents with DS included in this review are summarized in Table 2.

### Cardiovascular training

To the best of our knowledge, three studies have examined the effects of standardized aerobic training in children and/or adolescents with DS.

Varela et al. (2001) conducted a 16-week rowing ergometer training study, which was carried out on 16 adolescents and young adults with DS. Even though the exercise group achieved higher levels of work performance, no evidence of physical changes was found either in the body weight or in the percentage of body fat. Additionally, no changes in cardiovascular or physiological responses were found either in the treadmill test or in the rowing test. Similarly, Millar et al. (1993) designed a 10-week walking-jogging exercise program for 14 children and adolescents with DS, and yet again no changes were found in the cardiovascular capacities in any of the two groups, possibly due to the low exercise intensity; however, the exercise group showed an improvement in the time to exhaustion.

Ordoñez et al. (2006) focused on the aerobic training of 22 male adolescents with DS for 12 weeks, concluding at the end of the training period that they found a significant decrease in the percentage of fat mass (assessed by anthropometry) but reported no cardiovascular effects.

There have been several investigations only regarding the effects of training on body composition over a relatively short duration and the results of the studies

are nonconclusive due to the contradictory outcomes of each independent investigation. The studies have not shown improvements in cardiovascular fitness in the children and adolescents with DS, leading investigators to postulate that perhaps adaptations may require longer training periods and/or higher training intensities. New, specifically designed studies could contribute toward the validation of the hypothesis that cardiovascular capacity in children and adolescents with DS can be improved, as this has already been shown in adults with DS (Tsimaras et al., 2003).

### Strength training

Only one study was found which exercised youth with DS with a training program focused exclusively on strength. Weber and French (1988) studied a group of 14 adolescents with DS and designed two strength training programs: a weight training treatment and a strength exercise treatment. The participants performed 10 tests to evaluate their muscular strength before and after the treatment program. The results of this study were very clear and found that the group that performed the weight training program achieved significant improvement in muscular strength.

### Combined cardiovascular and strength training

In a case study by Lewis and Fragala-Pinkham (2005), a child with DS performed a 6-week home exercise program combining aerobic and strength training. After the training period, the results showed improvements in aerobic capacity and anaerobic power.

These studies related to training programs may pave the way toward to new research looking into increased strength levels that could have positive effects on health from several different approaches. For example, strength training could produce both neural and muscular-related strength increase and muscular hypertrophy, which in turn could reduce hypotonicity and balance dysfunctions and increase  $VO_{2max}$  and bone mass-related parameters. Because there are a number of significant benefits to be attained, efforts to elucidate the real effects of strength or cardiovascular-strength combined training should be promoted.

### Conclusions

Children and adolescents with DS are a unique population in relation to their health-related physical fitness variables. Body composition in this specific population is, in general, less healthy than that observed in their peers without DS, as proven by



higher BMI, lower levels of lean mass and reduced bone mass-related parameters.

Furthermore, children and adolescents with DS show lower levels of cardiovascular and strength capacities, which can result in a worse quality of life. Although there is a significant lack of information on youths with DS, it is evident that this population could benefit considerably from PA and exercise prescription. Data from the few studies available till now are contradictory in relation to improvements in body fat composition of the individuals. Adaptations have not been achieved in cardiovascular fitness when mild aerobic training is performed. One possible explanation for the lack of cardiovascular improvement may be a result of the low intensity and/or duration characteristics of the program described.

Further research in this topic would help to address pending issues such as the duration and intensity of aerobic training on improvement in cardiovascular fitness, or whether type, intensity and duration of strength training could be the most beneficial to children and adolescents with DS.

Importantly, the life expectancy of the population with DS is increasing with time; hence, cases of diseases some illnesses and diseases related to age

(until now relatively unreported for the DS population) such as osteoporosis or cellular aging begin to appear earlier than in the population without DS. Consequently, the main characteristics associated with DS can become more pronounced. The ultimate objective of future research in this field should be to test whether exercise (aerobic, strength and/or a combination of both) could benefit children and adolescents with DS, and help them have a healthier body composition and physical fitness, all of which result in a healthier and a better quality of life in this population.

**Key words:** exercise, body composition, cardiovascular fitness, aerobic, strength, training.

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