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BENEFITS OF PHYSICAL EXERCISE PROGRAMMES ON THE HEALTH OF INDIVIDUALS WITH DOWN SYNDROME: A SYSTEMATIC REVIEW

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ABSTRACT

Background: Down syndrome (DS) is the most common genetic cause of intellectual disability. Physical inactivity increases the risk of incidence of cardiovascular diseases, obesity and type II diabetes in individuals with DS. Regular physical activity is important for promoting their overall health and well-being across the lifespan. The aim of this systematic review was to synthesize the existing research concerning physical exercise and sports-based programmes for people with DS to determine their effects on the welfare of this population.

Methods: A systematic review was conducted by searching four electronic databases (Pubmed, SportDiscus, Cochrane and ERIC). Studies were included if they met inclusion criteria. The quality of the studies included in the review was assessed by the PEDro scale. All studies displayed a possible risk of bias. The information was extracted using the PICOT guidelines (Participants, Intervention, Comparison, Outcomes, Time).

Results: Thirteen studies met the inclusion criteria, of which ten explored the effects of a physical exercise programme whereas the other three explored the effects of a sports-based intervention. Improvements in lower and upper limb resistance, cardiorespiratory fitness, balance, walking capacity and body composition were found by these studies.

Conclusions: Physical activity has positive effects on the health of people with Down syndrome. It might be said that resistance and endurance are key components for a higher functionality and quality of life.

KEYWORDS: Down syndrome; physical exercise; health; quality of life; benefits/effects.

LABURPENA

Aurrekariak: Down sindromea adimen urritasunaren kausa genetiko ohikoena da. Jarduera fisiko ezak gaixotasun kardiobaskularrak, obesitatea eta II motako diabetesa pairatzeko arriskua areagotzen du Down sindromea duten pertsonengan. Bizitza osoan zehar jarduera fisikoa erregulartasunez egitea garrantzitsua da osasuna eta ongizatea sustatzeko. Errebisio sistematiko honen helburua ariketa fisikoko programekin zein kiroletan oinarritutako programekin erlazionatutako ebidentzia zientifikoa laburbiltzea izan zen, hauek populazio honen ongizatean duten eragina zehazteko.

Metodologia: Errebisio sistematikoaren bilaketa lau datu baseetan egin zen (Pubmed, SportDiscus, Cochrane and ERIC). Aukeraketa irizpideak betetzen zituzten artikuluak hartu ziren kontuan. Ikerketen kalitatea PEDro eskalaren bidez ebaluatu zen. Ikerketa guztiek alboratzeko arriskua egon zitekeela erakutsi zuten. Informazioa PICOT teknika erabiliz sailkatu zen, parte-hartzaileak, interbentzioa, alderaketa, emaitzak eta denbora kontuan hartuz.

Emaitzak: Hamahiru ikerketek betetzen zituzten ezarritako hautaketa irizpideak, hauetatik hamarrek ariketa fisiko programa baten efektuak aztertzen zituzten, eta beste hiruk, aldiz, kirol batean oinarritutako programa baten efektuak. Goiko nahiz beheko gorputz adarren indarrean, gaitasun kardiorrespiratorioan, orekan, oinez ibiltzeko gaitasunean eta gorputz konposizioan hobekuntzak eman zirela ikusi zen.

Konklusioak: Jarduera fisikoa praktikatzeak efektu positiboak ditu Down sindromea duten pertsonen osasunean. Esan liteke indarra eta gaitasun aerobikoa funtzionalitate eta bizi kalitate hobea izateko osagai gakoak direla.

HITZ GAKOAK: Down sindromea; ariketa fisikoa; osasuna; bizi kalitatea; onurak/efektuak

RESUMEN

Antecedentes: El síndrome de Down es la causa genética más común de discapacidad intelectual. La inactividad física aumenta el riesgo de padecer enfermedades cardiovasculares, obesidad o diabetes tipo II en personas con síndrome de Down. La práctica regular de actividad física es importante para mejorar su salud y bienestar a lo largo de la vida. El objetivo de esta revisión sistemática fue sintetizar la evidencia científica relacionada con diferentes programas de ejercicio físico e intervenciones vinculadas al deporte para personas con síndrome de Down, con el fin de determinar sus efectos en el bienestar de esta población.

Metodología: La revisión sistemática se llevó a cabo mediante la búsqueda en cuatro bases de datos (Pubmed, SportDiscus, Cochrane and ERIC). Los estudios fueron seleccionados si cumplían todos los criterios de inclusión fijados. Su calidad se evaluó mediante la escala de PEDro. Todos los estudios mostraron un posible riesgo de sesgo. La información fue extraída por medio de la técnica PICOT, que incluye participantes, intervención, comparación, resultados y tiempo.

Resultados: Trece estudios cumplieron con los criterios de inclusión, diez de ellos analizaron los efectos de un programa de ejercicio físico mientras que los tres restantes examinaron los efectos de las intervenciones basadas en deportes. Los estudios mostraron que se dieron mejoras en la fuerza tanto de extremidades inferiores como superiores, en la condición cardiorrespiratoria, en el equilibrio, en la capacidad de caminar y en la composición corporal.

Conclusiones: La actividad física tiene efectos positivos en la salud de las personas con síndrome de Down. Se puede observar que la fuerza y la capacidad aeróbica son componentes clave para tener una mayor funcionalidad y calidad de vida.

PALABRAS CLAVE: síndrome de Down; ejercicio físico; salud; calidad de vida; beneficios/efectos

INTRODUCTION

Down syndrome (DS), also known as Trisomy 21, is considered the most commonly known genetic cause of intellectual disability and it is also the most recurrent chromosomal abnormality in live births, with a frequency of one in approximately 700 births worldwide (Antonarakis, 2017; Bittles et al., 2007; Coppedè, 2016; Grieco et al., 2015; Mégarbané et al., 2009; Seron et al., 2017).

Approximately 150 years ago, in 1866, John Langdon Down was the first physician who described the phenotype of children with Trisomy 21 (Mégarbané et al., 2009). The discovery of karyotyping techniques in 1959 and their further development provided the knowledge that Trisomy 21 resulted from an additional chromosome in the 21^{st} pair of chromosomes (Mégarbané et al., 2009). Most cases of Trisomy 21 are due to meiotic nondisjunction, which consists in the failure of two members of a chromosome pair to separate properly during meiosis, producing a gamete with two copies of chromosome 21, instead of the single copy resulting from an ordinary meiosis (Coppedè, 2016; Sherman et al., 2007).

Many clinical disorders are attached to DS, including a high prevalence of cardiac, gastrointestinal, immunological, respiratory, sensory, and orthopaedic anomalies (Bittles et al., 2007). Besides these anomalies, other conditions, such as increased obesity, muscle hypotonicity, joint hypermobility, cardiac disease, hearing loss (Coppedè, 2016), vision disorders (Coppedè, 2016) and cognitive impairment (dementia, Autism Spectrum Disorder) (Grieco et al., 2015), have been associated with DS (Carmeli et al., 2002). These conditions may become more severe if the individual leads a sedentary lifestyle. Moreover, hypermobility of the joints was selected as a diagnostic sign of DS in neonates and children (Parker & James, 2008). Muscular hypotonia is the most regular anomaly of the neuromuscular system in babies with DS, which is a significant component that contributes to the previously mentioned joint hypermobility, and it is thought that the delay in the development of the cerebellum is associated with it. Hypotonia increases during early childhood and into the prepubertal period (Parker & James, 2008). People affected by DS suffer many age-related disorders earlier than the general population and a strong correlation has been found between the presence of congenital heart defects and death during the first 10 years of life (Bittles et al., 2007). Literature corroborates that adolescents with DS provide higher levels of overweight or obesity than other adolescents with or without intellectual disabilities at all (Suarez-Villadat et al., 2020). Decrease in flexibility is notable when fast growth associated with this period of life starts. This response may occur due to the fact that maturational changes and alterations happen in various tissues and in body composition (Parker & James, 2008). Small ears, flat face, flat nasal bridge, short neck, broad hands and a gap between the first and second toes are some of the physical characteristics individuals with DS have (Coppedè, 2016), whereas kindness, humour and forgiveness are some of their personality assets. It has also been observed that they show more positive facial expressions than typically developing peers (Grieco et al., 2015). Although cognitive development happens through childhood, adolescence and early adulthood (Grieco et al., 2015), the greater number of the population with DS (70–75%) reaches an intelligence quotient between 20 and 50 on average (Suarez-Villadat et al., 2020).

Some factors that may increase the risk of a DS pregnancy have been suggested by different authors (Coppedè, 2016; Morris, 2012). Most of the cases seem to be of maternal origin. The mother's advanced age at conception is known to be one of the major risk factors (Coppedè, 2016). Furthermore, other potential risk factors have also been investigated, for instance, maternal weight during pregnancy, the use of contraceptive pills, cigarette smoking and radiation exposure (Coppedè, 2016). Risk factors from paternal origin are less clear than those from maternal origin (Coppedè, 2016).

According to the analysis of the mortality data from the WA DSC (Western Australia, Disability Services Comission) cohort, the main causes of death in people with DS in the 19-40 year age group between 1953 and 2002 were pneumonia and other respiratory infections (23.1%), cardiac, renal, and respiratory failure (10.2%), cancers (7.7%), cerebrovascular accident (5.1%), and coronary artery disease (2.6%) (Bittles et al., 2007). Survival prospects for people with DS have dramatically improved in developed countries, from just 12 years in the 1940s to average 60 years nowadays (Bittles et al., 2007; Seron et al., 2017). Nonetheless, life expectancy is still lower than for the general population (Seron et al., 2017) and their quality of life deteriorates even further with age (Boer, 2020). Regular physical activity is important for promoting overall health and well-being across the lifespan. However, lack of physical activity raises the risk of obesity, type II diabetes and cardiovascular disease and may aggravate other fitness-related conditions associated with DS (Barr & Shields, 2011). Functional fitness main components include cardiorespiratory endurance, muscular strength and endurance, flexibility, balance, functional ability and appropriate body composition, which are key for the health and well-being of any population group. Reduced functional fitness hinders the ability to live a healthy and independent life (Boer, 2020). Improved well-being, increased self-esteem, prevention of chronic disease, rise of social interaction and a positive impact on their quality of life are some of the benefits that might be acquired by the regular practice of physical activity (Barr & Shields, 2011).

According to research, early intervention programmes can improve children's walking, balance and jumping skills, as well as, their level of intellectual functioning (Barr

& Shields, 2011). Engaging in regular physical activity may have a significant effect on individuals with DS's lifespan and their physical, mental and emotional health (Skiba et al., 2019), in addition to requiring less health intervention (Shields et al., 2013). In order to get those mentioned effects, adolescence is a strategic time. It is worthwhile to implement an exercise programme of good exercise habits early in life so that they can continue with healthy activity patterns in adulthood (Shields & Taylor, 2010). Even though there is much evidence of the benefits of having a healthy and physically active lifestyle, physical activity levels are extremely low in this population (Shields et al., 2013). The reasons for these low levels of physical activity are complex and multifactorial (Shields et al., 2013).

It is essential to encourage youth with Down syndrome to exercise on their way to adulthood as they become even less active during this period. As they are prone to chronic conditions such as diabetes, osteoporosis and obesity, exercising is especially important for them (Shields & Taylor, 2010; Shields et al., 2013). It has also been proved that a higher level of physical activity in individuals with DS means a reduced risk of incidence of cardiovascular diseases and helps increase their independence (Rimmer et al., 2004; Skiba et al., 2019), while a sedentary behaviour entails the opposite. People with DS experience an impairment of muscle strength, both in the upper and lower limbs of up to 50%, compared to other groups of the population without disability or with intellectual disability, other than DS (Shields et al., 2013). Children and young adults with DS show lower levels of cardiorespiratory fitness than their peers without DS, which can negatively affect their health and quality of life (Seron et al., 2017). Literature suggests significant positive differences in functional activities and work-related skills are related to the improvement of muscle strength (Shields et al., 2013). It is important to take into account that this population's jobs are usually associated with physical strength, so avoiding muscle weakness is crucial to be more efficient at the workplace. Furthermore, increasing lower limb resistance may help decrease the risk of falling down (Carmeli et al., 2002). Conversely, loss of independence, increased need for services and support and lower quality of life are some facts they can experience because of weak muscles and low aerobic capacity (Cowley et al., 2011). It is not certain the reason why low peak VO_2 values are found in this population, but according to several authors, low maximal heart rates (HR) and inactive lifestyles can contribute to it (Millar et al., 1992). Some physiological factors such as autonomic and metabolic dysfunction and reduced ventilatory capacity might be responsible for the previously mentioned low level of cardiorespiratory fitness (Seron et al., 2017).

Although there are currently no specific physical activity guidelines for children with DS, World Health Organization provides some recommendations for people with disability: children and adolescents are recommended to do at least physical activity (mostly aerobic) 60 minutes per day, as well as muscle strengthening exercises at least three times a week (World Health Organization, 2020). Recommendations concerning adults suggest doing "at least 150-300 minutes of moderate intensity or at least 75-150 minutes of vigorous-intensity aerobic physical activity throughout the week", and they should also strengthen major muscle groups twice or more times a week (World Health Organization, 2020). Lastly, older adults ought to do a multicomponent physical activity three times a week, in order to ameliorate functional capacity and prevent falls (World Health Organization, 2020).

It is necessary to consider the barriers and facilitators that individuals with DS may have when it comes to the practice of physical activity. On the one hand, according to research, four main facilitators have been found: family support, chance for social interaction with peers, structured appropriate programmes and the children's own physical ability and determination (Barr & Shields, 2011). On the other hand, four main barriers have been identified: characteristics directly linked to DS, family's priorities, either physical or behavioural poor skills and an absence of accessible programmes (Barr & Shields, 2011).

Therefore, the purpose of this review is to synthesize the existing research related to different physical exercise and sports-based programmes for people with DS to identify their effects on these people's health and well-being. The aggregate findings provide a comprehensive resource of current evidence that support physical activity providers in selecting exercise-based interventions, specifically for individuals with DS.

METHODS

This systematic review was reported using the PICOT guidelines. This structure consists of five components, successively, *Participants* (P), *Intervention* (I), *Comparisons* (C), *Outcomes* (O) and *Time* (T).

Eligibility criteria

To be included in this systematic review, studies needed to meet the following criteria: (a) full-text article published before November 2020; (b) participants with diagnosed Down Syndrome (both genders); (c) minimum sample of 10 people; (d) physical exercise related intervention; (e) pre-post-tests; (f) both control and experimental group; (g) outcomes referring to welfare; (h) interventions that lasted 8 weeks or longer; (i) at least total score of 5 points on the PEDro scale.

Dissertations, theses, conference proceedings, reviews and case studies were excluded. Studies involving participants with both Down syndrome and physical disabilities were also excluded.

Search strategy

Research articles were gathered using PUBMED, SPORTDiscus, COCHRANE, and ERIC database platforms, which represent databases from multiple disciplines related to health and physical activity. The search syntax included the following keywords with relevant Boolean operators inserted: “Down syndrome” AND “physical activity programme” AND “effects”; “Down syndrome” AND (“resistance training” OR “aerobic training”); “Down syndrome” AND (“aerobic training programme” OR “cardio* training programme” OR “resistance training programme”).

Studies were initially screened on the basis of title and abstract. Relevant abstracts were then selected for a full consideration of the article. Full articles were read in order to ensure the compliance of the inclusion criteria.

Study selection

The results from the searches were merged, and duplicate records of the same report were removed. Studies meeting the inclusion criteria were then separated into two sets, one focusing on resistance or/and aerobic training for people with DS and the other examining different sports-related interventions (swimming, Nordic walking).

Data abstraction

The abstracted data included bibliographic details of the papers (i.e., authors and year), participant characteristics (i.e., sample size, gender and average age), performed muscle group, intervention characteristics (i.e., type and duration) and outcomes.

Risk of Bias assessment of articles

To investigate the possible sources of bias the 11-item PEDro scale was used. This assessing instrument consisted of 11 items that could be answered with a “yes” or “no” format. The answer “yes” was awarded one point (if the criteria are satisfied) and the answer “no” was given zero points (if the criteria are not satisfied) for all items. Furthermore, each article was graded as ‘low’, ‘unclear’ or ‘high’ risk of bias for each domain.

The 11 items considered in PEDro scale were: whether “eligibility criteria were specified”; whether “subjects were randomly allocated to groups”; whether “the allocation of the groups was concealed”; whether concerning the most relevant prognostic indicators the groups were similar at baseline; whether “there was blinding of all subjects” (whether the subjects knew which group they had been allocated to); whether “there was blinding of all therapists who administered the therapy” (whether the therapists knew which group they were in charge of); whether “there was blinding of all assessors who measured at least one key outcome” (whether the assessors knew which group they were assessing); whether “measures of at least one key outcome were obtained from more than 85% of the subjects initially allocated to groups” (both the number of subjects initially allocated to groups and the number of subjects from whom key outcome measures were obtained); whether "all subjects for whom outcome measures were available received the treatment or control condition as allocated or, where this was not the case, data for at least one key outcome was analysed by *intention to treat*" (an intention to treat analysis means that where subjects did not receive treatment (or the control condition) as allocated, and where measures of outcomes were available, the analysis was performed as if subjects received the treatment (or control condition) they were allocated to); whether "the results of between-group statistical comparisons are reported for at least one key outcome"; whether "the study provides both point measures and measures of variability for at least one key outcome".

The studies were judged as (a) high risk of bias when the total score obtained on the PEDro scale was less than 4 out of 11, (b) unclear risk of bias when the total score obtained on the PEDro scale was between 4 and 7 out of 11, and (c) low risk of bias when the total score obtained on the PEDro scale was more than 7 out of 11. Articles graded as 'high' were then excluded.

Search results

Figure 1 presents the flow diagram. The initial search yielded 4700 results. Fifty-two duplicate studies were removed. Four thousand five hundred and nine records were removed because of other reasons. After removing duplicate records and other records for different reasons, as well as those not relevant to the topic of interest, 139 studies were screened. Furthermore, 107 studies were excluded based on their titles and/or abstracts. 32 full-text articles were assessed for eligibility, out of which 13 were retained in the systematic review.

A priori, only studies focusing on specific exercise programmes would be included in the review, such as resistance training programmes and aerobic training programmes. However, the intervention field was expanded including other types of intervention, for instance, Nordic walking training and swimming training programmes.

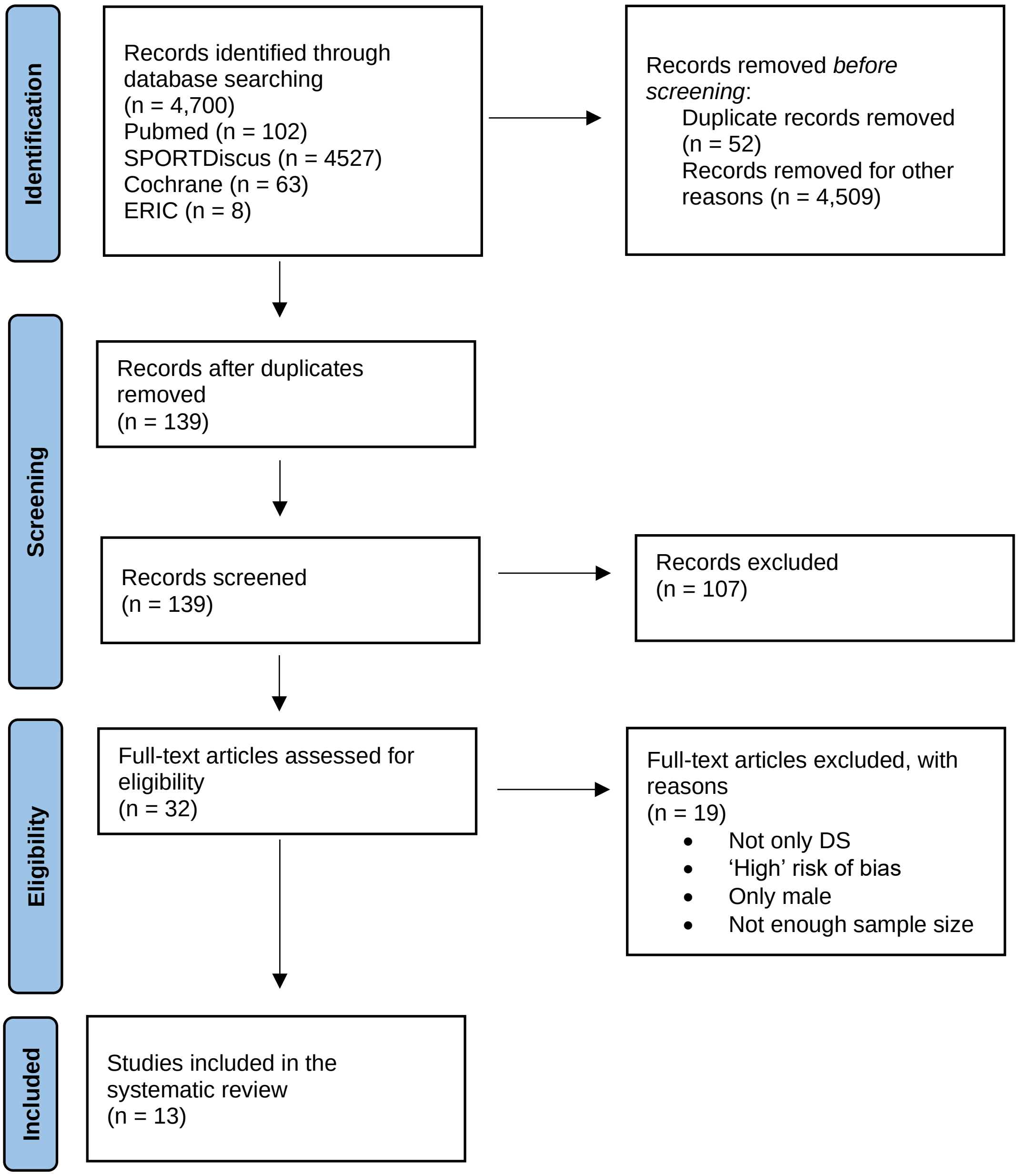

Figure 1. Flow diagram of the study retrieval process.

RESULTS

General results of the search

In this review studies meeting inclusion criteria were gathered. All these studies carried out physical activity programmes (detailed results are shown in Appendix). Three of the selected studies examined sports-related interventions (Boer, 2020; Skiba et al., 2019; Suarez-Villadat et al., 2020), whereas others performed physical exercise programmes (Alsakhawi & Elshafey, 2019; Carmeli et al., 2002; Cowley et al., 2011; Millar et al., 1992; Modesto et al., 2015; Rimmer et al., 2004; Seron et al., 2017; Shields et al., 2008; Shields & Taylor, 2010; Shields et al., 2013).

On the one hand, two out of the three sports-related interventions followed a swimming programme (Boer, 2020; Suarez-Villadat et al., 2020), while the other one followed a Nordic walking programme (Skiba et al., 2019). On the other hand, concerning physical exercise programmes, four studies followed progressive resistance training (Cowley et al., 2011; Shields et al., 2008; Shields & Taylor, 2010; Shields et al., 2013), two other studies completed an aerobic training session (Carmeli et al., 2002; Millar et al., 1992) and the remaining studies carried out both physical exercise types, merging resistance and aerobic training (Alsakhawi & Elshafey, 2019; Modesto et al., 2015; Rimmer et al., 2004; Seron et al., 2017). Moreover, one of those also added core stability training (Alsakhawi & Elshafey, 2019).

Sports-related interventions

As mentioned before, two interventions were related to swimming, where 71 individuals with DS took part in: 45 adolescents (Suarez-Villadat et al., 2020) and 26 adults (Boer, 2020). Adolescents followed a 36-week-duration front crawl and breaststroke training programme, whereas adults followed an 8-week-duration freestyle swimming programme. Both groups trained three times a week.

Although Suarez-Villadat, B. et al. (2020) found a significant decrease in all body composition variables for the experimental group (EG), they noticed an increase in their body composition variables for the control group (CG). There were significant differences in body mass index (BMI), waist circumference, waist-to-height-ratio (WtHR) and body fat. Moreover, significant differences in BMI, aerobic capacity, dynamic balance, resistance, 12-m swimming sprint and “8-ft get-up-and-go” between groups were found also in Boer’s (2020) study once intervention was finished.

22 adults with DS took part in Nordic walking intervention three times a week for 10 weeks (Skiba et al., 2019). Researchers did not find significant differences neither in spatiotemporal parameters nor in gait phases. Nevertheless, significant differences were found in Stance Phase (SP) in the right lower limb. They saw a desynchronised movement of the ankles joint compared to people without DS. In spite of the fact that

people of the EG improved synchronisation of the movement of the pelvis, the improvement was not statistically significant.

Progressive resistance training programmes

Within this type of training four studies were gathered (Cowley et al., 2011; Shields et al., 2008; Shields & Taylor, 2010; Shields et al., 2013). All of them followed a 10-week-duration programme, training twice a week. 141 people with DS took part in those programmes [91 adolescents/young adults (Shields & Taylor, 2010; Shields et al., 2013) and 50 adults (Cowley et al., 2011; Shields et al., 2008)].

Some studies show they improved lower and upper limb resistance after intervention (Shields et al., 2008; Shields & Taylor, 2010; Shields et al., 2013). However, the difference was not statistically significant neither for low (Shields et al., 2008), nor for upper limb (Shields & Taylor, 2010). Shields et al. (2013) found a significant difference in the "box stacking test" in EG at week 11 and a positive correlation between improved muscular strength and "box stacking test" and "weighted pail carry test" at week 24 for EG. In addition, Shields et al. (2013) also found an improvement in physical activity levels at week 24 for EG. In spite of those positive facts, they saw significant negative correlation between changes in upper limb resistance and changes in physical activity at week 11.

Furthermore, Cowley et al. (2011) discovered EG obtained better results in isokinetic strength in knee extensor and flexor peak torque, while they gained better results in isometric strength only in knee extensor peak torque. There was also an improvement in respiratory exchange ratio compared to CG. They found statistically significant positive changes in "ascend-descend 10 stairs test". However, improvement was not significant in "time to rise five times from a chair test".

Aerobic training programmes

40 people with DS took part in the two studies that analysed aerobic training programmes (Carmeli et al., 2002; Millar et al., 1992). 26 were older adults who trained for 25 weeks (Carmeli et al., 2002) and the other 14 were adolescents who trained for 10 weeks (Millar et al., 1992). Both studies' participants trained three times a week.

Even though both studies carried out the same type of aerobic training programme, each of them examined different variables. Besides improvement in walking duration, speed and distance, leg strength and balance also improved significantly in Carmeli et al. (2002) research for EG. "Timed-up and go" (TUAG) test showed better results after intervention too. During weeks 1 to 15 walking capacity (duration, speed and distance) increased, and then the parameters remained constant. Concerning the other aerobic training programme study (Millar et al., 1992), no significant differences were found in VO_{2peak}. However, treadmill test and minute ventilation, exercise heart rate (HR) and exercise duration were significantly improved.

Combined resistance and aerobic training programmes

Resistance and aerobic training programmes were performed in four studies (Alsakhawi & Elshafey, 2019; Modesto et al., 2015; Rimmer et al., 2004; Seron et al., 2017). Three of them (Modesto et al., 2015; Rimmer et al., 2004; Seron et al., 2017) followed a 12-week-duration programme training three times a week, while one (Alsakhawi & Elshafey, 2019) followed an 8-week-duration programme, also three times a week. Nevertheless, only one introduced both cardiovascular exercise and muscular strength for EG (Rimmer et al., 2004). Other three studies (Alsakhawi & Elshafey, 2019; Modesto et al., 2015; Seron et al., 2017) divided groups according to type of training (resistance training/ aerobic training). All in all, 179 individuals with DS took part in these studies (52 adults, 82 adolescents and 45 children).

Rimmer et al. (2004) discovered participants who followed the intervention improved cardiovascular fitness (VO_{2peak}14.1%, HR_{peak}, time to exhaustion and 27.1% maximum workload) and both lower and upper limb resistance (39%- 43%, respectively) compared to CG.

In two studies the participants were divided in three groups: control group (CG), aerobic training group (ATG) and resistance training group (RTG); one of them focused especially on cardiorespiratory fitness (Seron et al., 2017), whereas the other focused on resistance (Modesto et al., 2015). The study focusing on cardiorespiratory fitness found improvements for both ATG and RTG in VE_{max}. ATG increased total test time (TTT), but there was no significant difference in other groups. There was a VO_2 reduction in all submaximal stages, except for the 1st stage in RTG. Moreover, they found lower HR in ATG and RTG for submaximal stages, but there was no significant difference in HR for CG (Seron et al., 2017). Nonetheless, Modesto et al. (2015) found significant differences in body composition and maximum strength in ATG, whereas significant improvements were found just in maximum resistance for RTG. In addition, CG improved 1RM leg extension.

Finally, the study carried out by Alsakhawi & Elshafey (2019) was divided in three groups, respectively, traditional exercise programme (A), traditional exercise programme + treadmill training (B) and traditional exercise programme + core stability training (C). They found a significant improvement in functional balance and overall stability index for groups B and C compared to group A, but those differences were not statistically significant between groups B and C. These last two groups improved muscle strength after both training programmes, and consequently also balance.

DISCUSSION

Down syndrome (DS) is the most common genetic cause of intellectual disability (Antonarakis, 2017; Bittles et al., 2007; Coppedè, 2016; Grieco et al., 2015; Mégarbané et al., 2009; Seron et al., 2017). Health related comorbidities in these individuals are common and may be affected by a lack of active participation in physical exercise (Barr & Shields, 2011). The aim of this systematic review was to analyse the scientific evidence of the effects caused by different physical exercise and sports-based programmes for people with DS in order to know which are the benefits of each type of programme.

Sports-related interventions

Numerous sports-related interventions have been carried out with people with DS, but lack of homogeneity regarding the characteristics and types of intervention makes it difficult to compare them with each other because of several reasons. Firstly, even though all programmes are sports-related, they do not examine neither the same sport, nor the same variables. Secondly, the duration of the programmes varies much, from programmes that last 8 weeks (Boer, 2020) to programmes that last 36 weeks (Suarez-Villadat et al., 2020). Last but not least, the participants' age differs, from adolescents (Suarez-Villadat et al., 2020) to adults (Boer, 2020; Skiba et al., 2019). All these variations between studies do not allow to draw clear conclusions about which programme could be more efficient.

However, the decrease of most indicators of body composition after the-36-week-swimming programme strengthens the idea that high intensity training helps in the improvement of some indicators of body composition in people with DS (Suarez-Villadat et al., 2020). Another study related to swimming showed significant differences in BMI, aerobic capacity, dynamic balance, strength and 12-m swimming sprint among others (Boer, 2020). The increase in aerobic capacity is relevant since one of the risk factors for cardiovascular diseases in this population is low cardiorespiratory endurance (Boer, 2020). This increase together with the improvement in dynamic balance and resistance help to perform daily tasks better, which can increment the quality of life of these individuals significantly, due to the fact that these improvements may help to have more physical independence and less risk of falling, and as a result, more self-confidence. The improvement in the 12-m swimming sprint but not in the 24-m swimming sprint leads to consideration of the sort of training participants followed, given that they swam only one length at a time, so they did not train to turn efficiently. Therefore, it seems that despite the different duration, swimming programmes are efficient regarding the reduction of body composition.

Concerning the results of Nordic walking intervention, the short duration of the programme can be decisive to understand why Skiba et al. (2019) did not find many significant differences in some examined variables. It is also important to consider that changing the pattern of walking is not easy, and maybe a 10-week training programme is not enough to do it, because walking is an automatic process that is hard to modify (Skiba et al., 2019). Nevertheless, the improvement in the synchronisation of the movement of the pelvis may be due to the coordination needed between upper and lower limbs while practising Nordic walking.

Progressive resistance training programme

The results of the examined studies related to progressive resistance training programmes concurred investigating the effects of progressive resistance training for adolescents (Shields & Taylor, 2010; Shields et al., 2013) and adults (Cowley et al., 2011; Shields et al., 2008) with DS. All studies reported an improvement in lower and upper limb resistance after the programme, even if the changes were not statistically significant in all cases. This shows progressive resistance training is effective in order to increase both lower and upper limb muscle strength. However, it was surprising to find out that isometric knee flexor resistance did not change with training as Cowley et al. (2011) claimed. Whether the training programme was of sufficient intensity, frequency, and duration should also be taken into account. Apart from that, the participants of the studies that did not improve their lower or upper limb muscle performance may not have had the same potential as the ones who did. Using lower limb muscles to hold posture when carrying out daily life activities might explain the difference between upper and lower limbs, due to the fact that upper limb muscles are often less used (Cowley et al., 2011).

It cannot be said that the effect of progressive resistance training on the performance of work tasks is clear. Significant differences were noticed in the performance of the “box stacking test” in week 11, and positive correlation between improved resistance and “box stacking test” and “weighted pail carry test” were observed in week 24 (Shields et al., 2013). The specificity of training may also be pertinent. The “pail carry test” involves an isometric contraction of the upper limbs, but training consisted of isotonic contractions. Another parameter that this test assesses is mobility, which was not included in the exercises of the programme. Nevertheless, the “box stacking test” had more in common with the kind of training that was carried out. This could be one of the reasons why “box stacking test” reached significance after the programme. As changes in upper and lower limb resistance were linked to improvements in the performance of the “box stacking test” and “weighted pail carry test” may suggest that the duration of the programme was not long enough to carry over significant improvements in both tests. There is also a significant negative correlation between changes in upper limb muscle strength and changes in physical activity.

Physical activity levels had a significant difference after the programme carried out by Shields et al. (2013), at week 24. Perhaps people who took part in EG continued doing exercise on their own once the programme was finished, while people in CG did not. Another reason why this might have happened is the fact that the groups may not have been equal at baseline, but associations between changes in muscle strength and changes in physical activity do not support this idea.

As mentioned before, upper and lower limb resistance improved after the programmes (Cowley et al., 2011; Shields et al., 2008; Shields & Taylor, 2010; Shields et al., 2013). Concerning lower limb resistance, it is easy to believe that improving muscle strength may help to get better results in some aspects of daily life, such as ascending and descending stairs, and both speed and a lower degree of muscular effort while walking. Besides, improvements in upper limb resistance might be relevant in other aspects of daily life, for instance, reaching things from cupboards without help or also at work, which is important as most of the jobs individuals with DS do usually involve using these muscles. All these improvements help to enhance their quality of life.

In many cases, participants had a mentor with them while training. It seems this is a positive way to train for people with DS owing to the extra-motivation and higher demand these mentors can ask from them. In addition, this definitely increases confidence and it is easier for them to engage in exercise.

Aerobic training programmes

Even though both studies (Carmeli et al., 2002; Millar et al., 1992) carried out the same type of aerobic training programme, each of them examined different variables for different ages, which makes the comparison more difficult.

Nonetheless, scientific literature seems to show evidence that treadmill programs may improve leg strength in older adults (Carmeli et al., 2002), bringing with it improvements in the capacity of the subjects to keep balance. Moreover, this clearly supports the improvement in the TUAG test, as strengthening lower limb muscles and increasing balance facilitate standing up and walking faster, as it is also easier to control the movement. This fact may also be beneficial to decrease risk of falling.

Besides not finding significant differences in VO_{2peak} after 10 weeks of aerobic training, low cardiovascular fitness values were observed (Millar et al., 1992). It is surprising that participants did not improve VO_{2peak} although the training regime that they carried out was expected to produce improvements. There might be many factors that condition this finding. One of them could be the level of intellectual disability those participants have. In addition, it should be analysed if individuals with DS respond to training in an ordinary manner, due to the physiological limitations they have compared to their peers without DS. Although VO_{2peak} values did not improve significantly, other studied variables such as minute ventilation, exercise heart rate and exercise duration did, which means that the programme had positive effects on other aspects and it may decrease the risk of the presence of a cardiovascular event in the future.

Combined resistance and aerobic training programmes

Taking into account that resistance training is as beneficial as aerobic training for people with DS, it is interesting to analyse if combined training programmes are also effective for strengthening muscles, reducing body composition values and improving walking capacity and balance. A study that investigated the effects of cardiovascular fitness and both lower and upper limb resistance by the mentioned combined training programme found both parameters had improved (Rimmer et al., 2004). Even though more studies are necessary to confirm these findings, it is plausible that a combined training programme improves previously stated parameters. Moreover, it is widely believed that if individuals with DS improve muscular strength and endurance their functional decline may be delayed, maintaining their functional independence.

Another study compared the effectiveness of treadmill training exercises and core stability exercises in children with DS (Alsakhawi & Elshafey, 2019), it concluded that both types of training improved muscle strength, and so did balance. It is believed that both training programmes have similar effects on gait patterns by positive changes in static and dynamic balance. However, longer duration could cause wider differences between treadmill and core stability exercise programmes.

Concerning VO_{2peak} values, Seron et al.'s (2017) results concurred with previously mentioned findings of Millar et al. (1992), suggesting that these values did not improve after the intervention. It should be considered if the intensity of the exercises was enough in order to increase VO_{2peak} and what low HR_{peak} values entail, apart from the possible reasons explained above. Furthermore, increasing pulmonary ventilation might be beneficial in individuals with DS due to the fact that some of their physical characteristics include restricted size of airways and small nasal passage, which can interfere during exercise. Even though improvements in VO_{2peak} were not achieved, total test time (TTT) was improved by the aerobic training group (ATG) after the programme (Seron et al., 2017), which suggests that participants could continue to exercise for a longer time until exhaustion. Lower HR values were also found, which means that training had made their heart more efficient, as the same intensity rate requires less work. Consequently, these two findings (improved TTT and lower HR values) could be considered as a positive adaptation and might help in their everyday life.

The decrease obtained in body composition after doing the aerobic training (Modesto et al., 2015) also concurred with the results of other studies carrying out swimming programmes (Boer, 2020; Suarez-Villadat et al., 2020). However, that decrease was not observed in the group that followed the resistance training. This may lead to two (among others) possible conclusions: firstly, that the intensity of both training programmes was different or secondly, that aerobic training is more efficient in order to decrease body composition variables.

Limitations of the study

A limitation of the current review is the large amount of heterogeneity that was presented across studies because in spite of the fact that participants carried out the same type of programme, the measured variables were different. Moreover, the relatively small samples of the studies hinder generalization of the obtained results for all the individuals with DS. Another limitation is that the short duration of the programmes and their frequency prevented some expected improvements for some variables. Finally, despite carefully devising search keywords and strategies, some potential literature may have been excluded.

Future observations

It would be interesting to study the benefits that longer duration programmes could have and the long-term sustainability they have in order to evaluate whether it is necessary to carry on with this kind of-programme or if it is not worth it. It would also be interesting to know how the improvements obtained with the training programmes could alter their mental health and their daily life.

CONCLUSION

Based on this review, physical exercise programmes and sports-related interventions have a strong evidence base to support their effectiveness. Improvements in aerobic capacity may facilitate daily life activities for individuals with DS. Besides, increasing lower limb resistance helps to ameliorate balance, which is important in order to prevent falls. On the other hand, gaining muscle strength in upper limbs is also beneficial because their jobs often include using these muscles. It has been proved that aerobic training programmes and sports-based programmes are useful for positive alterations in body composition. Resistance and endurance are key components for the health and well-being of this population, due to the fact that their functionality and quality of life rise by the improvement of the mentioned components. Finally, it must be said that it is difficult to specify which type of programme is the most effective in improving the welfare of these people as all the programmes showed positive results.

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APPENDIX

	Authors (year)	Sample	Quality (PEDro scale)	Target muscles	Intervention	Variables	Results
1	Alsakhawi, R.S., Elshafey, M.A. (2019)	n= 45 (4-6 years; 15A, 15B, 15C) Children with DS	8	CORE, lower and upper limbs	Group A (CG): traditional exercise programme to improve posture control and balance (60') Group B: same programme as group A for 30' in addition to treadmill training. Three times a week for 8 weeks. Group C: same programme as group A for 30' in addition to core stability training. Three times a week for 8 weeks.	Functional balance (Berg Balance Scale) Dynamic balance (Biodex Balance System)	Significant improvement in functional balance and overall stability index in groups B and C compared to group A. No significant differences between group B and C in functional balance and overall stability index. Improvement in muscle strength, and consequently also in balance after both training programmes (B and C).
2	Boer, P. H. (2020)	n= 26 (13 female, 13 male; 18-45 years; 13 EG, 13 CG) Adults with DS	8	Full body	EG: freestyle swimming training, resting after each length. W 1 to 4 - 30', W 5 to 8 - 40'. Three times a week for 8 weeks. CG: no structured physical activity	11 functional fitness tests: Balance (static (standing on one leg) + dynamic (walking on balance beam)), flexibility (shoulder stretch), functional fitness tests ("6-min walk distance" + "8-ft up and go"), muscular strength and endurance tests ("sit-to-stand", modified curl-up + isometric push-up), aerobic test ("16-m PACER" and speed (12- and 24-m swimming sprint tests).	Significant differences in BMI, aerobic capacity, dynamic balance, resistance, 12-m swimming sprint and "8-ft get-up-and-go" between groups once intervention was finished.

3	Carmeli, E., Kessel, S., Coleman, R., Ayalon, M. (2002)	n= 26 (16 female, 10 male; 57-65 years; 16 EG, 10CG) Adults with DS	7	Lower limb	EG: training in treadmill at 0% incline 10'-15', gradually increasing time up to 45'. Three times a week for 25 weeks, CG: no change in their levels of physical activity	Isokinetic muscle strength (Biodex dynamometer) Dynamic balance and gait speed (TUAG - "timed-up and go" (modification of "get-up and go"))	EG's speed improvement in the TUAG test compared to the first testing. Improvements in walking duration (+%150), speed (+%86) and distance (+%180). Significant improvements in leg strength and balance. During weeks 1 to 15 walking capacity (duration, speed and distance) was increased, then the parameters remained constant.
4	Cowley, P. M., Ploutz-Snyder, L. L., Baynard, T., Heffernan, K. S., Jae, S. Y., Hsu, S., Lee, M., Pitetti, K. H., Reiman, M. P., & Fernhall, B. (2011)	n = 30 (13 female, 17 male; 20-38 years; 19 EG, 11 CG) Adults with DS	5	Lower and upper limbs	EG: progressive resistance training. 7 exercises, increasing workload during the programme. (8-10 x 3). Twice a week for 10 weeks. CG: regular activities	Isometric and isokinetic knee extensor and flexor strength (Biodex System 3 dynamometer) Peak aerobic capacity (treadmill, individualised protocol) Functional tasks of daily living (time to rise five times from a chair, walk 25ft (7,62m) + ascend-descend 10 stairs.	EG's improvement in knee extensor and flexor peak torque for isokinetic strength compared to CG. EG's improvement in knee extensor peak torque, but not knee flexor peak torque for isometric strength compared to CG. EG's improvement in "ascend-descend 10 stairs test" but no significant improvements in "time to rise five times from a chair". CG's increase in peak relative VO_2 compared to EG. EG's increase in respiratory exchange ratio compared to CG.
5	Millar, A. L., Fernhall, B., Burkett, L. N. (1992)	n=14 (3 female, 11 male; 15-22 years; 10 EG, 4 CG) Adolescents with DS	6	Lower limb	EG: 30' of continuous brisk walking and jogging. Three times a week for 10 weeks. CG: no regular physical training	Maximal oxygen consumption (modified Balke protocol in treadmill)	No significant differences in VO_2 peak after intervention. However, significant improvements in minute ventilation, exercise heart rate and exercise duration. EG's significant improvement in treadmill test compared to CG.

6	Modesto, E. L., de Almeida, E. W., Carani, I. G., Greguol, M. (2015)	n= 41 (16 female, 25 male; 12-20 years; 16 ATG, 15 RTG, 10 CG) Adolescents with DS	5	CORE, lower and upper limbs	ATG: aerobic training (HRR 50-70%). Three times a week (50') for 12 weeks. RTG: resistance training. 9 exercises (2x12 / 45"). Twice a week (50') for 12 weeks. CG: no training	Muscle maximum strength (1RM high row + 1RM leg extension) Handgrip strength (dynamometer JAMAR, isometric contraction during 5") Body composition	ATG significant differences in body composition and maximum strength. RTG significant improvements in maximum strength. CG improvement in 1RM leg extension.
7	Rimmer, J. H., Heller, T., Wang, E., Valerio, I. (2004)	n= 52 (29 female, 23 male; 30-70 years; 30 EG, 22 CG) Adults with DS	8	Lower and upper limbs	EG: 30'-45' cardiovascular exercise and 15'-20' muscular strength and endurance (initiated at 1RM 70%, then weight increased by 10% of their 1RM). Three days a week for 12 weeks. CG: no regular physical training	VO_2 peak Resistance (1RM bench press + 1RM seated leg press) Handgrip strength (Grip-A handgrip dynamometer)	EG's improvement in cardiovascular fitness (VO_{2peak} (14.1%), HR_{peak}, time to exhaustion + maximum workload (27.1%)) and strength (both lower and upper limb, 39%-43%) compared to CG.
8	Seron, B. B., Modesto, E., Stanganelli, L. C. R., Carvalho, E. M. O. D., Greguol, M. (2017)	n= 41 (16 female, 25 male; 12-20 years; 16 ATG, 15 RTG, 10 CG) Adolescents with DS	5	CORE, lower and upper limbs	ATG: aerobic training. Treadmill + stationary bike (15' each, HRR 50-70%). Three times a week (50') for 12 weeks. RTG: resistance training. 9 exercises (3x12 / 1')3'. Three times a week (50') for 12 weeks. CG: no physical activity	Cardiorespiratory fitness (VO_{2peak}, VE_{max} + HR_{peak}), on the treadmill.	CG VO2peak significantly lower in W12. ATG and RTG improvements in VE_{max}. ATG's increase in TTT (total test time), no significant differences in other groups. VO_2 reduction in all submaximal stages, except for the 1st stage RTG. Lower VE values for ATG and CG. ATG and RTG lower HR in submaximal stages, not significant differences in HR for CG.

9	Shields, N., & Taylor, N. F. (2010)	n= 23 (6 female, 17 male; 13-18 years; 11 EG, 12 CG) Adolescents with DS	9	Lower and upper limbs	EG: progressive resistance training. 6 exercises, increasing workload during the programme (3 x 12/2'). Twice a week for 10 weeks. CG: regular activities (leisure + sporting activities)	Muscle strength (1RM chest press + leg press) Lower limb physical function (functionality) ("Timed up and Down Stairs test") Upper limb physical function (functionality) ("Grocery Shelving Task test")	A 42% increase in lower limb muscle strength compared to CG. No significant differences between the groups for upper limb muscle strength or upper and lower limb physical function.
10	Shields, N., Taylor, N. F., & Dodd, K. J. 2008	n= 20 (7 female, 13 male; 20-49 years; 9 EG, 11 CG) Adults with DS	9	Lower and upper limbs	EG: progressive resistance training. 6 exercises, increasing workload during the programme (2-3 x 10-12/2´). Twice a week for 10 weeks. CG: regular activities	Muscle performance (1RM seated chest press + 1RM seated leg press) Muscle endurance Physical function ("timed stairs test" + "grocery shelving task")	EG's improvement in weight lifted during training in the 6 exercises. Statistically significant improvement in upper limb muscle endurance, muscle strength and functionality compared to CG. No significant differences between two groups in lower limb muscle endurance, muscle strength and functionality compared to CG.
11	Shields, N., Taylor, N.F., Wee, E., Wollersheim, D., O'Shea, S.D., Fernhall, B. (2013)	n= 68 (30 female, 38 male; 14-22 years; 34 EG, 34 CG) Young adults with DS	9	CORE, lower and upper limbs	EG: progressive resistance training. 7 exercises, increasing workload during the programme. (3 x 12/2'; 1RM %60-80). Twice a week (45'-60') for 10 weeks. CG: social programme (arts + recreational activities). Once a week (90') for 10 weeks. Measurements W0, W11 and W24.	Work task performance ("weighted box stacking test" + "weighted pail carry test"), muscle strength (1RM single seated chest press + 1RM seated leg press) and physical activity levels (RT3 activity monitor).	A 21% increase in upper limb resistance and a 30% increase in lower limb resistance (-) compared to CG. Significant difference in the box stacking test in EG (W11). Significant negative correlation between change in upper limb resistance and change in physical activity (W11). Improvement of physical activity levels (W24). Positive correlation between changes in upper and lower limb resistance and performance of the box stacking test (W24). Also positive correlation between changes in upper limb resistance and performance of the weighted pail carry test (W24).

12	Skiba, A., Marchewka, J., Podsiadlo, S., Sulowska, I., Chwala, W., & Marchewka, A. (2019)	n= 22 (11 female, 11 male; 25-40 years; 11 EG, 11 CG) Adults with DS	5	Full body, mainly lower limb	EG: Nordic walking. Three times a week (60') for 10 weeks. CG: no training during intervention	Gait, based on the changes of the spatiotemporal parameters (step length, cycle length, speed, standardized step length + standardized speed) and the peak values of the angles of the selected joints, compared to people without DS.	No significant differences in spatiotemporal parameters. Desynchronised movement of the ankles joint compared to people without DS. Significant differences in SP in right lower limb. No significant differences in gait phases. Improvement in synchronisation of the movement of the pelvis, but not statistically significant.
13	Suarez-Villadat, B., Luna-Olivac, L., Acebesa, C., Villagra, A. (2020)	n= 45 (20 female; 25 male; 12-15 years; 15 EG, 30 CG) Adolescents with DS	7	Full body	EG: front crawl style and breaststroke style training, using technical support elements. Intensity, volume, types of loads and recovery time was changed during the programme. Three times a week (50') for 36 weeks. CG: water games (recreational), swimming 300-400m on average per session. Twice a week for 36 weeks.	BMI Waist circumference (WC) WtHR (WC + equally fair for short and tall persons) %BF Skinfold measurements	EG's significant decrease in all body composition variables. CG's increase in all body composition variables. Significant differences in BMI, waist circumference, WtHR and body fat between groups.

DS: Down syndrome; W: Week; CG: Control Group; EG: Experimental Group; ATG: Aerobic Training Group; RTG: Resistance Training Group; SP: Stance Phase; WtHR: Waist-to-Height-Ratio