

# **A SYSTEMATIC REVIEW ON EFFECTS OF PHYSICAL ACTIVITY INTERVENTIONS ON EARLY MOTOR DEVELOPMENT IN CHILDREN WITH DOWN SYNDROME**

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**Abstract**

**Purpose**

The main aim of this review was to evaluate the effect of physical activity on motor development in children with Down Syndrome by means of a systematic review.

**Methods**

An extensive literature search of published studies in English from 1980 to May 2006 was performed. Of the fifty-eight studies identified only four met the inclusion criteria. This review included studies that investigated the effects of physical activity on motor development in children with Down Syndrome and evaluated the outcomes in terms of the level of activity.

**Results**

One study showed a significant decrease in length of time to independent walking in the intervention group (CI -101[-180.48 - -21.52]. Two studies (N=84) reported a significant improvement in the total developmental quotient following intensive physical activity (wmd and CI 95% -13.07 [-17.66, -8.48]. Three studies showed an increased in locomotor developmental skills following physical activity intervention.

**Conclusion**

The results of this review support the use of programmes that are designed to improve motor development in children with Down Syndrome.

We recommend that physical activity programmes need to be intensive and parents should be incorporated to strengthen the outcomes.

**Key words**

Down Syndrome, physical activity, systematic review, motor development.

## Introduction

Down syndrome is a genetic disorder occurring in one out of 800 births and is strongly associated with mental retardation, memory and speech problems, limited vocabulary and slow motor development (Spiker & Hopmann, 1997; Chapman & Hesketh, 2000). It is observable at birth and in 95% of cases and is usually due to the result of failure of the twenty-first pair of chromosomes to separate during meiosis resulting in the individual inheriting three of these chromosomes rather than the normal two (Berk, 2004). Chromosome pair 21 is the smallest of the 23 human chromosome pairs, possessing only about 1.5% of the total genetic material (Cicchetti & Beeghly, 1990). In addition to the common features associated with the syndrome does the affected person also suffer from mild to moderate mental retardation and delayed motor development. Early visual and auditory information-processing difficulties have been recognized as causes for interference with learning and acquisition of basic developmental skills (Cicchetti & Beeghly, 1990).

Fine motor and cognitive skills go hand in hand during the early stages of development, through manipulating objects in the child's environment (Bruni, 1998). A developmental approach according to Cicchetti & Beeghly (1990), suggests that the objectives of intervention should focus on improving optimal functioning in basic areas of development such as motor, cognitive, language, social-emotional development etc (Cicchetti & Beeghly, 1990). Investigators who have adopted a developmental perspective have demonstrated that the course and content of symbolic play development in children with Down Syndrome is markedly similar to that observed in normal children (Cicchetti & Beeghly, 1990).

Development of gross motor skills can be particularly rewarding, as it is an area where progress results are easier to be observed (Winders, 1997). Hypotonia in children with Down Syndrome affects their motor development. It is most easily observed when they are in infancy (Winders, 1997). Children with Down Syndrome have increased flexibility in their joints because the ligaments that hold the bones together have more slack than usual resulting in hyperextension (Bruni, 1998; Winders, 1997). Because of the laxity, children with Down Syndrome are more prone to dislocations. Children with Down Syndrome have muscle weakness but strength can be greatly improved through repetition and practice (Winders, 1997). Studies related to Down Syndrome mainly apply the developmental perspective and examine the interaction between heredity and environment on the developing organism (Bruni, 1998).

Physical activity has been widely used in the treatment of children with motor impairments. Several studies have documented effects of physical activity on motor development in children with cerebral palsy and developmental delay (Palmer, 1997; Condon, 2002). Physical activity is a broad term that encompasses all forms of exercise or movement that can range from sports to lifestyle activities. In Kanda, Pidcock, Hayakawa, Yamori & Shikata (2004) four of five children who completed physical activity training could either stand still for five seconds or walk at the time of the outcome evaluation 52 months after the beginning of the therapy program. None of the five subjects with no training or insufficient training could accomplish this task when evaluated 64 months following therapy initiation. Although the number was small, the difference was found to be statistically significant ( $P=0.0278$ ). In another study of twenty-

nine children with meningomyelocele (MMC) and shunted hydrocephalus, all had motor impairment, but after physiotherapy and training, walking was possible in 23 of them (5 autonomously and 18 with an aid), while six had recourse to a wheelchair (Rendeli, et al, 2002). A statistically significant cognitive level was also found after the intervention between the ambulatory clients (both with and without aids) and those who were dependent on wheelchairs (P Intellectual Quotient: 83-85 vs 63).

Lee & Smith found a 69% improvement in motor function and skills following individual physiotherapy sessions with children (Lee & Smith, 1998). Three months after the commencement of the treatment, there was a 73% improvement especially in relation to the gross motor abilities, self-confidence and social skills.

It has however been previously documented that NDT does not have significant effects in children's neurological development (Lilly, Powell, 1990). These results could not be generalized as the study had only two participants. A few other studies have failed to find convincing evidence for the efficacy of physical therapy on therapeutic early intervention for infants (Turnbull, 1993; Palmer, Shapiro, Wachtel, Allen, Hiller, Harryman, et al 1988; Goodman, Rothberg, Houston-McMillan, Cooper, Cartwright & van der Velde, 1985).

Although physical activity interventions for Down Syndrome are increasing, current literature is unclear on the effects of physical activity on motor development in children with Down Syndrome. A systematic review of evidence addressing motor developmental outcomes of physical activity is required.

The main aim of this review was to evaluate the effect of physical activity on motor development in children with Down Syndrome aged of ten years and below by means of a systematic review.

## **Methods**

### **Literature search**

An extensive literature search of published studies in English from 1980 to May 2006 was performed using the key word for Down Syndrome including, Down Syndrome and Trisomy 21 combined with search strategy for Intervention, motor development, physical activity, physical therapy, structured play etc. Sources for relevant studies included databases such as, Ebscohost, MEDLINE, ERIC, CINAHL, MEDLINE, Infotrac and hand searches of referenced articles from obtained articles as well as links from searched electronic articles. Two independent assessors (OK and CVN) who reviewed the trials using a standardized protocol did the inclusion of articles, based on title and abstract.

### **Selection criteria**

The inclusion criteria were that the study should investigate the effects of physical activity on motor development in children with Down Syndrome aged ten years and below and evaluate outcomes in terms of the level of activity and participation. Intervention was at least bi-weekly for a minimum period of 12 weeks or longer. Any intensive physical activity or stimulation as well as any neuro-developmental therapy could be used as the intervention. Studies included in this systematic review were all those written in English with prospective randomized controlled research designs including quasi-randomized studies, using comparative groups. Studies had to have a comparative group on non-treatment or ordinary treatment without intensive physical activity. Any standardized / validated scale

such as the Bayley Scale or Griffith's scale could be used to measure the outcome (Griffiths, 1996; Bayley, 1993). Outcomes include total developmental quotients, motor development quotients, motor development measures and measures of performance.

### **Data extraction**

The methodological validity of all searched studies was reviewed by two reviewers (OK and CVN). The authors of the trials were known to the reviewers. The two reviewers extracted data from the studies that fulfilled the inclusion criteria using a standardized extraction form. In case of incongruity the reviewers resolved it through discussion. Eight studies met the general inclusion and exclusion criteria. Only three studies had data available in a format to use for this review. Two were randomized controlled trials and one had a quasi-experimental design.

### **Data analysis**

All data obtained was continuous data. The following variants are acknowledged: Age difference of participants, different interventions and different measurement instruments. These variants make synthesis of data extremely difficult and results should be interpreted with caution. A statistician from the Medical Research Council (Cape Town, South Africa) was consulted to look at the available data from the studies and confirmed that a meta-analysis is difficult under these circumstances and recommended that subgroup analyses would be more appropriate. The authors therefore commented on individual study

outcomes in subgroup analyses and used the combined meta-analyses with caution. The design, methodological quality, type of outcome measures and statistical significance of the results are taken into consideration in the synthesis.

The included studies were assessed by two independent reviewers (OK and CN) to measure the methodological quality of the included studies using the PEDro scale. The PEDro scale is an 11-item scale designed for rating methodological quality of RCTs (Maher, Sherrington, Herbert, Moseley & Elkins, 2003). The first item on the scale measures external validity and the other ten measure internal validity (Pang, Eng, Dawson, Gylfadottir, 2006). It has been reported that the PEDro scale provides a more comprehensive measure of methodological quality (Bhogal, Teasell, Foley & Speechley, 2005). The first item has a yes and no response and seeks to identify if the study report describes the source of the participants and has a list of criteria used to determine who was eligible to participate in the study. The other ten items are used to calculate the PEDro scale (partitioned) score and measure internal validity. They have a one-point score each and therefore the highest score is ten points. The higher the score, the better the quality of the randomized controlled trial. A score of nine to ten is rated as excellent; six to eight, good; four to five, fair and less than four, poor. A point is awarded only in a case where the study clearly mentioned that the criterion was met. According to Table 1, two studies scored 6, and two scored 5 respectively.

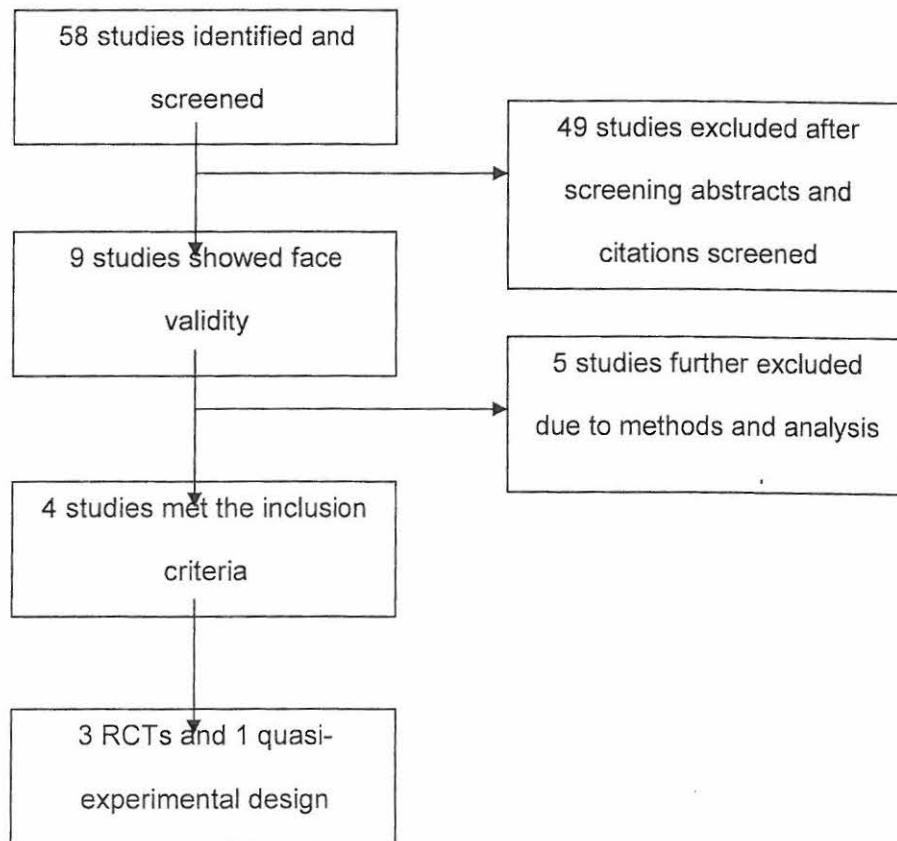
**Table 1 Methodological quality scores on the PEDro Scale**

Study ID	External Validity	Internal Validity	Score
Giudice	Yes	RA, BS, ITA, SC, PM/MV	5
Piper	Yes	RA, BS, MO, SC, PM/MV	5
Ulrich	Yes	RA, BS, MO, ITA, SC, PM/MV	6
Uyanik	Yes	RA, BS, MO, ITA, SC, PM/MV	6

RA – Random Allocation, CA – Concealed Allocation, BS – Baseline Similarity, PB – Participant Blinded, TB – Therapist Blinded, AB – Assessor Blinded, MO – Measures of key Outcomes from more than 85% of Participants, ITA – Intention to Treat Analysis, SC – Between Groups Statistical Comparisons, PM/MV – Point Measures and Measures of Validity.

During the search 58 studies were identified that met our initial query for inclusion. Forty-nine studies did not meet the further criteria of being randomized controlled trials and were excluded. Nine trials were identified that made use of randomization and one used a quasi-experimental design (Figure 1).

**Figure 1 Flow diagram of included studies**



### **Excluded studies**

Data were unavailable in one trial (Connolly, Morgan & Russell, 1984) and another compared Down Syndrome children with normal children (Connolly, Morgan, Russell & 1980). Mayo (1991) included children with cerebral palsy in the trial. Connolly and Michael (1986) never reported on motor development and Mahoney, Robinson and Fewell (2001) did not record relevant data for analysis. For these reasons the studies were not included. Assessments were done between three and 12 months. The analysis was done based on intention to treat.

### **Included studies**

Guidice used a computer-generated randomization to allocate 47 children with Down Syndrome to the experimental and comparison groups (Guidice, Brogna, Romano, Paludetto, Toscano, 2006). The experimental group underwent a parent-implemented developmental training programme by means of the Carolina Curriculum for infants and Toddlers with Special Needs (CCITSN). The parents conducted the interventions at least twice a day between meetings with tutoring professionals (Table 2). The comparison group received the standard therapist implemented treatment provided by the National Health Service of the Italian Region of Campania (NHST).

The aim of this study was to assess whether parent implemented developmental training using CCITSN could be of greater benefit to young children with Down Syndrome than the standard therapist implemented treatment provided by the NHST. Thirty-two infants with Down Syndrome completed the study. Children in the intervention group received developmental training at home twice daily with parents (n=21). Children in the NHST group were attended to at NHS rehabilitation centers by physiotherapists weekly (n=11). Therapy sessions

lasted 50 minutes and were carried out three times for each child weekly. Neurodevelopmental Therapy was the main used technique by the therapists. The outcome measure was the developmental quotient (DQ).

In Ulrich, Ulrich, Angulo-Kinzler and Yun (2001) 30 infants with Down Syndrome were randomly allocated to treadmill walking (intervention group, n=15) or control group (n=15). The purpose of the study was to determine if practice stepping on a motorized treadmill could help reduce the delay in walking onset normally experienced by these infants. Infants received traditional physical therapy at least every other week. In addition the intervention group received practice stepping on a small motorized treadmill, five days per week, for eight minutes a day in their own homes. Parents administered the treadmill intervention. The mean age of the infants was 307.4 days. The interventions were done till the child could walk. The 34th item on the Bayley Scales of Infant Development (2nd Edition) was administered. The primary outcome measure was the length of time from entry into study to onset of walking independently. Another study was designed to stimulate normal development in children with Down Syndrome aged less than twenty-four months (Piper, Pless, 1980). Thirty-seven participants were allocated to the treatment group (n=21) or the control group (n=16). The control group received no intervention. The intervention programme consisted of one hour duration, bi-weekly sessions over a period of six months. Parents were given a set of written instructions to follow at home between sessions. The Griffith's Scales of Mental Development were used to assess changes in the developmental status in the two groups (Griffiths, 1990).

Uyanik Bumin and kayihan (2003) randomly allocated forty-five children with Down Syndrome to three intervention groups; sensory integrative therapy (SIT, n=15), vestibular stimulation (VS) in addition to sensory integrative therapy (n=15) and Neurodevelopmental therapy (NDT, n=15). Each

session for all the participants lasted for one hour and a half, three days per week over a period of three months Locomotor skills were measured by 10 step forward walking and 10 step side walking. A home programme was given to all participants.

**Table 2 Characteristics of the included studies**

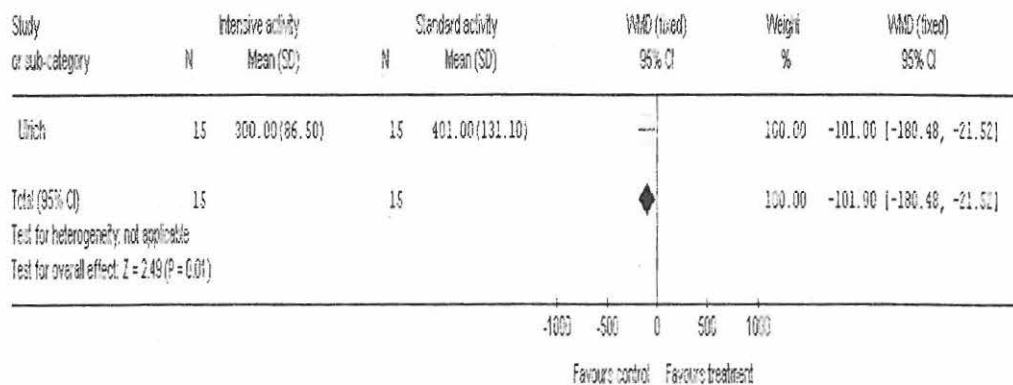
Study ID	Methods	Participants	Intervention	Outcome
Giudice	Computer-generated Randomised	47 Infants with DS 32 were analysed after fallout, Average age (CCITSN, 4.5 months), (NHST, 5.9 months)	Parent-implemented developmental training programme at least twice daily (CCITSN, n=21) versus standard therapist implemented treatment (NHST, n=11), NDT	Developmental quotient Gross motor skills Fine motor skills
Ulrich	Randomised Allocation	30 Infants with DS, No fallout mean age – 307.4 days	Treadmill Training for 8 minutes, 5 days/week (n=15) versus standard treatment (n=15). All participant (N=30) received at least bi-weekly physical therapy sessions	Independent walking Locomotor skills
Piper	Quasi-experimental	37 infants with DS Less than 24 months old No fallout	Treatment group (n=21), stimulation of normal development for an hour, bi-weekly for 6 months verses Control group, no treatment (n=16)	Developmental quotient Locomotor developmental quotient
Uyanik	Randomised Allocation	45 children with DS 7 to 10 years No fallout	SIT, n=15 VS 1 SIT n=15 NDT, n=15	Locomotor skills

We acknowledge that there were different interventions and different measures used in the studies. Comparison groups were also different.

There was only one study that measured length of time to independent walking.

**Figure 2 Effect size for length of time to independent walking**

Review: A Systematic Review on Effects of Physical Activity Interventions on Early Motor Development in Children with Down Syndrome (Version 1)  
 Comparison: 01 Intensive physical activity vs standard treatment  
 Outcome: 01 Length of time to independent walking (Days)



This study show a significant decrease in length of time to independent walking in the intervention group (CI -101[-180.48 - -21.52]. Although it is a single study with a small number of participants, we

conclude that the intensive physical activity intervention significantly decreased time to walking (Figure 2).

Figure 3 Effects sizes for improvement in development quotient

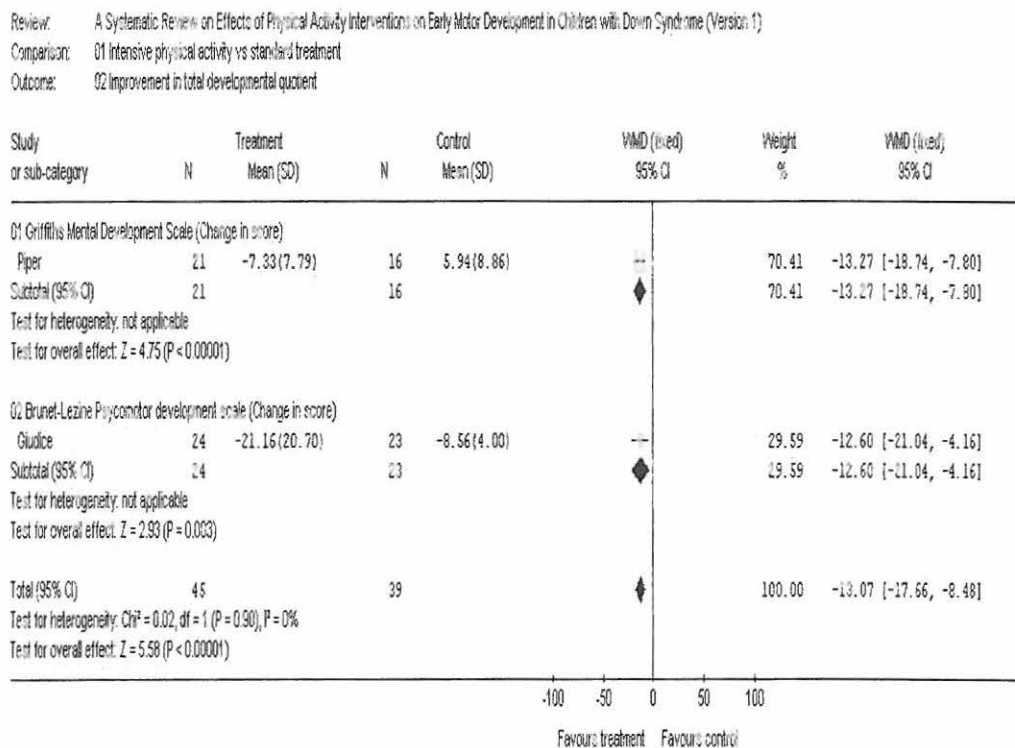


Figure 3 shows that the two studies (N=84) reported a significant improvement in the total developmental quotient following intensive physical activity (wmd and CI 95% -13.07 [-17.66, -8.48]. In Piper and Pless (1980) a bi-weekly physical activity intervention over a period of six months designed to stimulate normal development produced a significant increase on the developmental quotient with an effect estimate of wmd= -13.27 (95% CI, -18.74, -7.80).

significant increase in the development quotient in participants that participated in the programme (Giudice et al, 2006). It had an effect size of wmd = -12.60 (95% CI, -21.04, -4.16). The study reported a significant improvement in the developmental quotient scores of children that received a developmental training over a 12-month period compared to a comparison group. Parents were used in the intervention.

Another programme called the Carolina Curriculum for Infants and Toddlers with Special Needs (CCITSN) that consisted of developmental training produced a



**Figure 4 Effect sizes for locomotor developmental skills**

Review: A Systematic Review on Effects of Physical Activity Interventions on Early Motor Development in Children with Down Syndrome (Version 1)  
 Comparison: 01 Intensive physical activity vs standard treatment  
 Outcome: 03 Improvement in Locomotor Developmental Skills

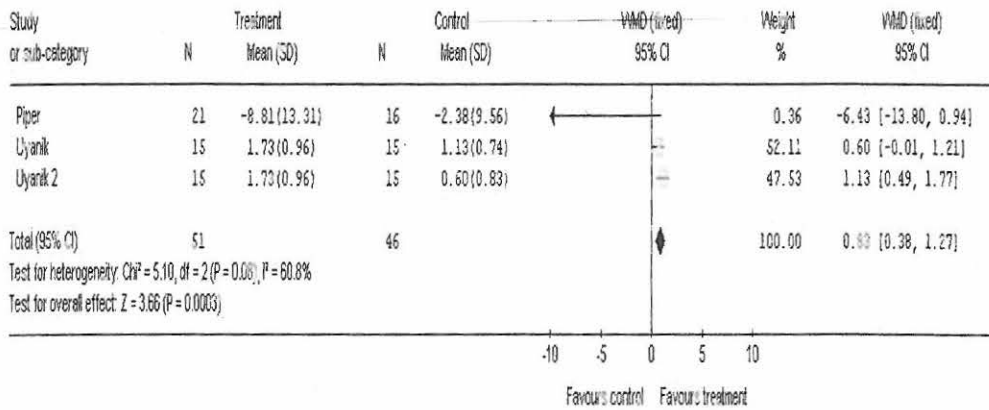


Figure 4 displays the effect sizes for changes in development quotient of locomotor developmental skills following an intensive physical activity programme. The three studies as shown above shows that physical activity produced increased locomotor developmental skills in children with Down Syndrome with a total effect size of  $wmd=0.83$  (95% CI, 0.38, 1.27). Two studies in Uyanik et al (2003) showed increase in locomotor developmental skills, with effect sizes of  $wmd = 0.60$  (95% CI, -0.01, 1.21) and  $wmd = 1.13$  (95% CI, 0.49, 1.77). Uyanik looked at locomotor development and the sub-studies are referred to as Uyanik a and Uyanik b.

**Discussion**

The purpose of this study was to determine the effectiveness of physical activity on motor development in children with Down Syndrome. The inclusion criteria required that an intensive physical activity programme be administered to the children for

at least a bi-weekly interval over three months. Only three studies met the inclusion criteria. Exclusion of studies ensures that results represent the capabilities of physical activity to influence motor development.

Several studies that evaluated the effects of physical activity on motor development were excluded. For example, Connolly and Michael (1986) reported on motor skills in the intervention group but never reported the same for the comparison group. Most of the studies reported interventions that had an NDT component.

Blinding of the participants as well as the therapist was not common. It is not feasible in many cases where the therapist has to supervise the intervention. Two studies showed improvement in motor development following physical activity interventions and one didn't. The one that didn't show a significant improvement had a less intensive regimen compared to the other two.

Studies using parents as interventionists have shown better outcomes (Ulrich et al, 2001; Giudice et al, 2006). Many researchers have used parents in the implementation of physical activity programmes. A commitment to using parents as interventionists is the most effective and cost-efficient way of providing services to young children with Down Syndrome and other developmental disabilities (Piper & Bless, 1980).

Research should use more standardized measures in studies investigating the effects of physical activity interventions for children with Down Syndrome.

### Conclusion

The results of this review support the use of programmes that are designed to improve motor development in children with Down Syndrome. Intensive physical intervention significantly shortens the time that Down Syndrome children would normally take to walk independently. Furthermore does intensive physical therapy increase total and locomotor development quotient significantly. We recommend that physical activity programmes need to be intensive and parents should be incorporated to strengthen the outcomes. It is further recommended that researchers should carry out studies of higher quality to provide better evidence on the effectiveness of physical activity on motor development in children with Down Syndrome.

### Limitation of the systematic review

Blinding is not possible in an intervention such as intensive physical activity is investigated and the possibility of bias may influence the assessor. The studies included in this review are of fair quality. We acknowledge that it is difficult to meta analyze results when the intervention and the scale of measure differs.

### Implications for research and clinical practice

It is recommended that researchers should embark on larger randomized control trials using similar assessment instruments and similar intensive physical intervention activities. There is limited literature on physical activity interventions on children with Down Syndrome.

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