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The Role of Antigravity Treadmill in Haemophilic Children Aged 8 to 11 Years with Unilateral Knee Haemarthrosis without Radiological Involvement

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Author's contribution

The sole author designed, analyzed and interpreted and prepared the manuscript.

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ABSTRACT

Background: Recurrent joint bleeding in children with hemophilia leads to joint damage with pain, loss of range of motion and function. Antigravity treadmill (AGT) allows unloading of the lower extremities during exercise in a pressurized treadmill chamber.
Aim: To investigate the effect of AGT on muscle strength and functional balance in children with hemophilic knee arthritis.
Study Design: Prospective, randomized controlled study.
Place and Duration of Study: The outpatient clinic, Medical rehabilitation hospital, and King Fahd Hospital, Almadinah Almonawarah, Kingdom of Saudi Arabia, between January 2015 and Mars 2015.
Methods: Thirty hemophilic male children with unilateral knee heamarthrosis in the age ranged from 8 to 11 years old were randomly allocated into two equal groups. The control group (group A) received traditional exercise program, while the study group (group B) received AGT gait training added to the same exercise program of group A. Training was applied three times per week for twelve consecutive weeks. All children were assessed at baseline and after 36-session.

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Assessment included peak torque of quadriceps and hamstring muscles using Biodex Isokinetic Dynamometer and functional balance using Pediatric Berg balance scale (PBBS).

Results: Significant differences were observed in both groups when comparing their pre and post treatment mean values of all measuring variables ($p<0.05$). Peak torque of the quadriceps changed from (32.60±1.183, 32.933±1.533) to (38.40±1.681, 40.466±1.060) and peak torque of the hamstring changed from (25.00±1.81265, 25.933±2.0517) to (32.533±2.474, 35.933±2.631) for the control and study group respectively, indicating more improvement in favor of the study group. Both groups demonstrated a significant increase in functional balance, with more improvement in favor of group B.

Conclusion: It can be concluded that gait training using AGT may be used as a therapeutic intervention for improving muscle performance and functional balance in children with hemophilic knee arthritis. Although our sample was not large, the effect size was moderate to large (0.85 and 0.83) for the knee extensors and flexors respectively and 0.553 for PBBS.

Keywords: Anti-gravity treadmill; isokinetic; peak torque; knee heamarthrosis; balance.

1. INTRODUCTION

Hemophilia is a bleeding disorder that occurs primarily in males and is *inherited* in an *X*-linked *recessive* pattern. It is characterized by a deficiency in one of the clotting factors, which hinders the normal cessation of bleeding in these patients [1]. Hemophilia is classified into two types: hemophilia A (Factor VIII *deficiency*) and hemophilia B (deficiency of Factor IX) [2].

The most frequent site of serious bleeding in patients with hemophilia is the knee joint because it is the largest, most exposed, and most vulnerable joint to sport or work injuries during everyday life [3].

Recurrent bleeding into the joints in hemophilic patients leads to joint damage with resultant contractures, joint deformities and arthritis. This in turn leads to muscle weakness and atrophy, limited participation in physical activity, osteoporosis and disability [4].

The decrease of muscular strength in hemophiliacs has been showed by different authors, who have measured muscular strength in hemophiliacs compared with control subjects [5,6]. Involvement in appropriate physical activity is an important component of the management of hemophilia [7].

Sensory input from vestibular, visual and proprioceptive systems is necessary to control posture and balance. Impairment in proprioception due to repetitive joints bleeding may lead to a deficit in postural balance which, in turn, leads to high joint stress and risk of falling [8]. In a study performed by Souza et al. [8] twenty children with haemophilia and 20 age-matched children control were recruited. A force plate was used to record variation in the centre of pressure (COP) displacement during the following conditions: standing on firm surface with eyes open, standing on foam surface with eyes open, standing on firm surface with eyes closed and standing on a foam surface with yes closed. Variables of COP as mean velocity and sway area and in anterior-posterior and medio-lateral direction were processed and for each variable sensory, quotients were calculated in both groups. Hemophilic group demonstrated a higher value in sway area variable on proprioception quotient when compared with the control group.

Several types of rehabilitation techniques have been used for children with hemophilia. Treadmill walking with body weight support can decrease weight bearing on the lower extremities and is frequently used for patients with musculoskeletal and neurological impairments. In particular, AGT is effective for decreasing ground reaction forces and provides safe gait training after surgical procedures [9]. However, the effects of AGT on the hemophilic knee arthritis children are unknown. The aim of this study was to investigate the effect of AGT training on muscle strength and functional balance in children with hemophilic knee arthritis.

2. SUBJECTS MATERIALS AND METHODS

2.1 Subjects

Thirty volunteer hemophilic male children (type A and B) with unilateral knee hemarthrosis, their ages ranged from 8 to 11 years participated in this study. They were selected from the outpatient clinic, Medical rehabilitation hospital, and King Fahd Hospital, Almadinah Almonawarah, Kingdom of Saudi Arabia. They were treated with factor replacement therapy (on-demand) therapy for bleeding events. They were divided randomly into two groups of equal numbers, control (group A) and study (group B), 15 patients for each. They were selected according to the following criteria: (1) Age of patients ranged from 8 to 11, years, (2) They suffered from unilateral knee hemarthrosis, (3) Pain and bleeding ranged from mild to moderate according to the classification of hemophilia recommended by the Orthopedic Advisory Committee of the World Federation of hemophilia [10], (4) They were able to walk independently; (5) All children were clinically and medically stable, (6) They were not suffering from acute joint and muscle bleeds during treatment time and (7) They were in pre-radiological phase of haemophilic arthropathy .

The exclusion criteria included: (1) children with advanced radiographic changes including (bone destruction, bony ankylosis, knee joint subluxation or epiphyseal fracture), (2) congenital or acquired skeletal deformities in both lower limb and (3) surgical procedures performed six weeks prior to treatment or during the exercise program.

Approval of the study was achieved from the Institutional Review Board of Medical rehabilitation hospital, and King Fahd Hospital. Written informed consent was obtained from parents or legal guardian, and additional verbal assent was obtained from those children who were willing to participate.

2.1.1 Randomization

Following the baseline measurements randomization process was performed using closed envelopes. The investigator prepared 30 closed envelopes with each envelope containing a card labeled with either control group or study group. Finally, each child was asked to draw a closed envelope that contained whether he was allocated to the control group or study group.

Both groups received exercise therapy program including stretching and strengthening exercises for one hour 3days/week, while the study group; received ,in addition, gait training using AGT for 20 minutes.

2.2 Materials

2.2.1 Materials for evaluation

2.2.1.1 Tanita HD-384 Digital Weight Scale

It was used to record patients 'weight.

2.2.1.2 The Biodex Isokinetic Dynamometer

Biodex medical system, Shirley, New York, USA was used for assessment of isokinetic muscular performance of the quadriceps and hamstring muscles of all participants. It is one of the most comprehensive computers driven, biomechanical musculoskeletal assessment and rehabilitation with 3 pro multi-joint systems. Trial-to-trial and day-to-day reliability and validity torque measurement of the Biodex system 3 were all previously established [11].

2.2.1.3 The pediatric Berg balance scale

This scale includes 14 items that assess daily functional activities of children from 5 to 15 years with mild to moderate motor impairment. This scale has demonstrated reliability and ease of application [12].

2.2.2 Materials for treatment

The AGT consists of a standard treadmill covered with inflatable fabric enclosure that covers the treadmill. In the center of the enclosure is a hole through which the user steps onto the surface of the treadmill. The subject puts on a special pair of shorts, and these shorts are zipped into the hole in the enclosure. The enclosure is filled with air by a blower. The pressure inside the inflated enclosure provides a lifting force against the body. The AGT offers a level of comfort while exercising unmatched by other unweighting systems.The percentage of body weight supported is determined by the air pressure in the bag, which provides a lifting force on the body [13].

2.3 Methods

2.3.1 For evaluation

2.3.1.1 Muscle torque

Isokinetic pre-test and post test measurements of concentric hamstrings and quadriceps muscles were performed for every child at velocity of (60 deg/sec) using Biodex Isokinetic dynamometer.

Prior to the evaluation, the participants performed warm-up exercise on ergometric bicycle for 5 minutes and stretching of quadriceps and hamstring, in a single set of 30 seconds for each muscle group.

The child was sat on the dynamometer chair according to the manufacturer's recommendations and was suitably stabilized with belts around his thorax, hip and thigh of the tested limb to avoid compensations. All testing began with the subject's knee at 90° of knee flexion. For the quadriceps test, the subjects concentrically extended their knee to full extension (0°).For the hamstring test, the subject concentrically flexed the knee back to 90°. The rotation axis of the dynamometer was aligned with the rotation axis of the tested knee joint (lateral femoral condyle), to avoid that the torque measurements were invalidated. Assessment was carried out by the same evaluator, who encouraged the child during the entire test with verbal commands to perform his maximal strength.

During isokinetic evaluation and as complement to the warm-up, the subjects performed a familiarization session on the dynamometer at the same velocity used in the test in order to reduce the learning effects and guarantee the reproducibility of the collected data.

2.3.1.2 Functional balance

It was assessed using the PBBS, which consists of 14 items and is scored using a 5-point Likert scale from 0 to 4, with zero denoting an inability to perform the activity without assistance and four indicating the ability to perform the task with complete independence. The score is based on the time for which a position can be maintained, the distance to which the upper limb is capable of reaching in front of the body, and the time required to complete the task. The maximum score is 56 points. The test is performed with the child clothed and making use of his/her habitual brace and/or gait-assistance device [14,15].

2.3.2 Methods of treatment

2.3.2.1 Control group

The control group received a designed exercise program for one hour, it was conducted 3 days/week for 3 successive months in the form of strengthening exercises and stretching exercises.

Stretching exercises: were conducted to maintain length of calf, hamstring, hip flexors and adductors of lower limbs and wrists, fingers flexors and elbows flexors as follows: The extremity was moved slowly through the free range to the point of restriction. Padding was used in areas where there is minimal subcutaneous tissue or bony prominence. Firm and comfortable grasp was applied proximal to the joint to firmly stabilize the proximal segment and distal to the joint to move the distal segment. Very gentle traction force was applied to the moving joint in order to avoid joint damage during stretching procedure. The stretching force was applied in a gentle, slow, and sustained manner by taking the joint to the point of tightness and then moved just beyond without inducing pain. Stretching was applied for 30 seconds followed 30 seconds rest and repeated 5 times/day and 3 sessions/week. This time for rest between sessions allows tissue healing and minimizes post exercise soreness.

Strengthening exercises: Static muscle contraction for quadriceps, hamstrings, dorsiflexors and planter flexors for 15 min. Each child performed five times initially, building up to 10 repetitions as tolerated, two to three times per day [16].

2.3.2.2 Study group

The Alter G Anti-Gravity treadmill (AGT) was used for gait training. It consisted of a treadmill that was enclosed in a pressurized bag. Gait training program was performed for 20 min, 3 times per week for 12 weeks. The exercise session passed through the following steps:(1) Putting on the shorts: The child wore a pair of neoprene shorts, (2) stepping into the AGT :the cockpit was lowered so it compressed the bag against the treadmill surface then the child entered from the back and stepped into the opening in the fabric enclosure, (3) Adjusting the height of the cockpit, 4) zipping into the fabric enclosure: neoprene shorts were zipped into the bag, (5) Adjusting percentage of body weight with the + and – button controls, body weight support was set at 30% of the child's body weight (6) speed adjustment: Speed was increased and decreased by pressing the + or – button controls. The practice on the treadmill was done at 75% of over-ground speed and zero degree inclination based on the work of others [17,18].

2.3.3 Statistical analysis

Data were managed and analyzed using the Statistical Package for Social Sciences (SPSS version 16.0). Descriptive statistics were calculated to summarize the data set for both groups and to identify potential baseline differences between the groups; p values were used to indicate the strength of the evidence. P value less or equal to 0.05 was considered significant.

A mixed between-within subjects analysis of variance was conducted to compare scores on the mean values of peak torque of the knee extensors and flexors at (60 deg/sec) of both groups A and B across 2 time periods: (pre- intervention), and (post -intervention) with the within-subjects factor being time and the between-subjects factor being treatment method.

For the pediatric Berg balance scale. Comparison between the two groups was performed using Mann–Whitney U test while comparison between variables within the same group was performed using Wilcoxon test. The effect size for quantifying the effectiveness of the two interventions (traditional or AGT) was calculated.

3. RESULTS

3.1 Patient Characteristics

The characteristics of the participants are shown in Table 1. The two groups were matched for age (p=0.827) and body weight (p=0.520). The distribution of hemophilia A and B in both groups was different. Also the distribution of factor deficiency was different in both groups. After random assignment of subjects to both groups, the distribution of hemophilia A and B in the control group and the study groups was 11/4 and 12/3 respectively. Also the distribution of factor deficiency was 0/ 11/ 4 and 0/ 12 /3 respectively; but before the baseline evaluation one subject with hemophilia B and mild factor deficiency decided to withdraw from the study and was replaced with another subject with hemophilia A and moderate factor deficiency then re-randomization was performed.

3.2 Peak Torque

Regarding to peak torque of the knee extensors at 60 deg/sec, there was significant interaction between type of intervention (traditional or AGT) and time, Wilks' Lambda =0 .806, F (1, 28) = 6.734, p 0.015. There was a significant main effect for time, Wilks' Lambda = .076, F (1, 28) = 340.255, p <0 .001, partial eta squared =0 .974, with both group subjects showing an increase in mean values of peak torque of the knee extensors at (60 deg/sec) of groups (A and B) across the two time points Table 2 and Fig. 1. The main effect comparing the two groups of subjects was significant, F (1, 28) = 10.230, p =0 .003, partial eta squared =0 .268, suggesting significant difference in mean values of peak torque of the knee extensors scores between group A and group B in favor of group B. The mean difference for the study group was 7.53 and the percentage of improvement was 22.874.

Concerning peak torque of the knee flexors at 60 deg/sec, there was significant interaction between type of intervention (traditional or AGT) and time, Wilks' Lambda =0 .616, F (1, 28) = 17.461, p (< .001). There was a significant main effect for time, Wilks' Lambda =0 .026, F (1, 28) = 1.033, p <0 .001, partial eta squared =0 .974, with both group subjects showing an increase in mean values of peak torque of the knee flexors at (60 deg/sec) of groups (A and B) across the two time points Table 2 and Fig. 2.

The main effect comparing the two groups of subjects was significant, F (1, 28) = 6.537, p = .015, partial eta squared = 0.194, suggesting significant difference in peak torque of the knee flexors scores between group A and group B in favor of group B. The mean difference for the study group was 10.00 and the percentage of improvement was 38.561.

Table 1. Characteristics of subjects in both groups

	Control group	Study group
Age in years(Mean±SD)	9.354±0.882	9.426±0.8957
Weight (kg) (Mean±SD)	28.112±1.163	27.856±0.910
Height (cm) (Mean±SD)	129±2.56	127.3±3.12
Haemophilia A/B	12/3	12/3
Factor deficiency (severe/moderate/mild)	0/ 12 / 3	0/ 12 / 3

Table 2. Mean ±SD of pre and post treatment mean values of peak torque (Newton. meter) of the knee flexors and extensors (60 degrees/ sec) for both groups.

	Pre intervention n= 15	Post intervention n= 15	$F_{(1,28)}$	P
Knee extension				
Control group	32.60±1.183	38.40±1.681	10.230	<0.05
Study group	32.933±1.533	40.466±1.060		
Knee flexion				
Control group	25.00±.1.81265	32.533±2.474	10.881	<0.05
Study group	25.931±.2.05171	35.933±2.631		

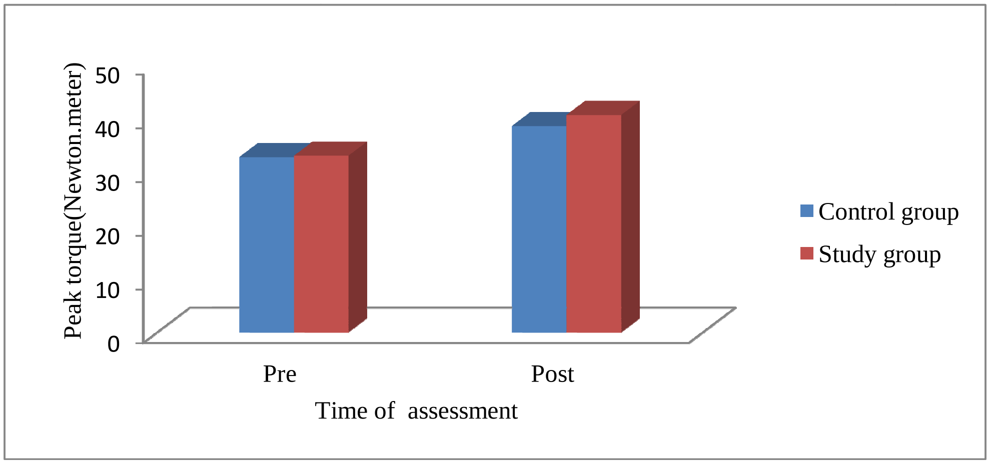

Fig. 1. Pre and post intervention mean values of peak torque of the knee extensors at (60 deg/sec) of both groups (A and B)

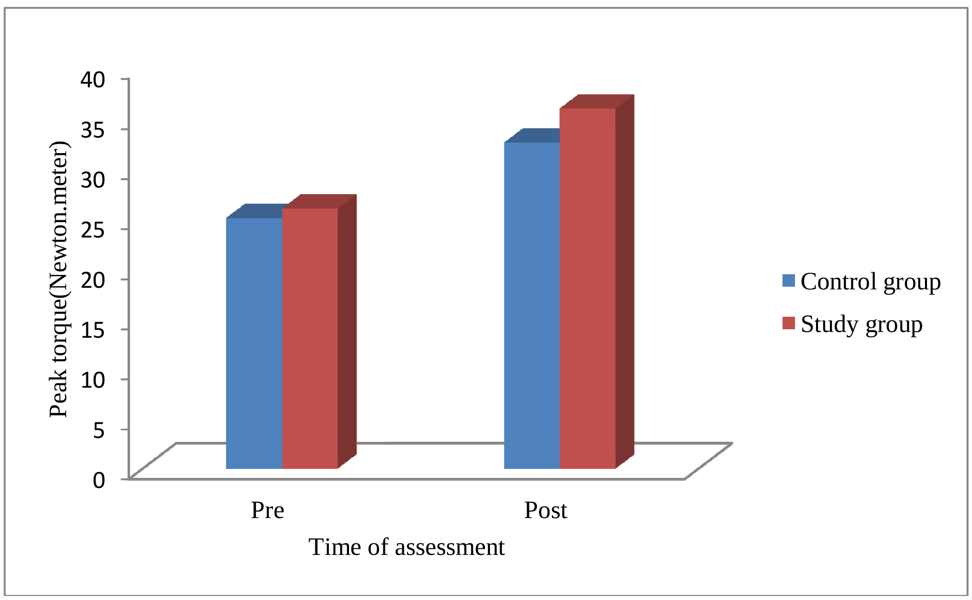

Fig. 2. Pre and post intervention mean values of peak torque of the knee flexors at (60 deg/sec) of both groups (A and B)

3.3 Functional Balance

The within group analysis demonstrated that both groups achieved higher scores on the Berg balance scale following both protocols. However, the between group analysis demonstrated that the group B had statistically better results in comparison to the group A (p=0.01) Table 3.

4. DISCUSSION

This article demonstrates that AGT training can be used with positive outcomes in children with hemophilia, with ages ranging from 8 to 11 years, to improve muscle strength and balance. The improvement in the study group may be attributed to the following factors: effect of AGT on reduction of energy expenditure during walking; decrease the muscular work performed to overcome internal friction within the joint; reduction of pain; sensory integration ; decrease ground reaction force and feeling of safety while exercising on the AGT.

Many previous studies showed that, when humans walk at normal weight, metabolic requirement increases with velocity [19-21]. The greater metabolic demand of faster walking may be attributed to increases in stride frequency, increases in mechanical power, and generation of greater ground reaction force (GRF) over shorter periods of ground contact [22-24]. Other studies have shown that when a portion of body weight is supported using a weight suspension system, less metabolic power is required to walk [25,26].

This reduction of metabolic energy while walking using AGT helped the child to conserve energy that he uses for postural control. During gait, these children consume more mechanical energy. A great amount of this energy is used to maintain balance. Stiffening the body through co-contractions could be one of the strategies used to maintain balance. As these co-contractions from antagonist muscles do not produce any movement, they consume a lot of metabolic energy with no apparent work produced. Part of the muscular work may also be used to overcome internal friction and viscosity in the ligaments, muscles, or joints in order to deform the body segments. This work is supposed to be greater in patients with hemophilia as joint stiffness and muscle fibrosis are common complications of arthropathy [27].

AGT is effective in improving balance as it allowed the child to walk while the upper extremities are free to move; which in turn challenged the child's balance. This Comes in agreement with Kurz et al. [28] who stated that AGT is effective treatment for improving walking biomechanics and balance of children with motor disabilities. The AGT offered other advantages over conventionally used harness systems and was very well accepted by the children [28].

Also, our results come in agreement with Martin et al. [29] who investigated the functional effects of combined aerobic and strength training in patients with Becker and limb-girdle muscular dystrophies. Eight patients performed 10 weeks of aerobic and strength training on AGT. Significant improvement was observed in dynamic postural balance that was assessed 10 weeks prior to training, immediately before training and after 10 weeks of training.

AGT has been proven to be effective in reducing pain during gait after anterior cruciate ligament reconstruction and meniscetomy, It is a useful mean of graded rehabilitation and early partial weight-bearing through unloading of the lower extremities [30]. This reduction of pain, during walking, allowed the child to concentrate in one task. This concentration might have improved the integration of different sensory information to have a better balance and postural control through providing gradual challenge to children to improve their motor abilities to meet the demands of motor control.

Table 3. Comparison of pre and post intervention mean values of Pediatric Berg Balance Scale scores of both groups

	Pre intervention (Mean±SD)	Post intervention (Mean±SD)	Z	P
Group A	46.60±0.9856	48.866±0.9154	-3.372	0.001*
Group B	46.733±0.883	50.200±1.08233	-3.508	0.000*
U	104	9		
P value	0.711	0.000*		

**:Significant*

AGT preserves many of the natural biomechanical patterns in ambulation, providing an advantage over previous unweighting systems. Research has demonstrated that AGT can be used to significantly decrease ground reaction forces at the knee joint, while maintaining normal patterns of muscle activation, joint range of motion, limb swing mechanics and cardiovascular function during walking [30-32]. All the previously mentioned factors helped children to walk without fatigue which in turn improved muscle strength.

In a patient with hemophilia, muscle imbalance may occur as a direct result of bleeds or indirectly from poor posture, and insufficient flexibility. Insufficient attention to this aspect of rehabilitation may lead to recurrence of symptoms [33]. The increase in muscle strength, recorded in this study, may be due to providing support and confidence to start walking and take more steps in comfort. This in turn prolonged the duration of walking which in turn gave the participants a way to build endurance and leg muscle strength without the usual pounding and impact lower extremities [34].

The increase in muscle strength observed in our study comes in agreement with Eastlack et al. [30] who reported improvement in muscle contraction in patients who performed AGT exercise after knee surgery.

Also, our results come in agreement with Koch and his colleagues [35]. In their research about the effect of strength training program over patients with hemophilia reported significant increase in muscles strength of these patients. They stated that the increasing power and range of motion, was associated with reducing the times of bleeding. Although the training methods of subjects were different in our study, the results correspond to Koch and his colleagues findings. The feeling of safety while exercising on the AGT may be a contributing factor in the improvement in muscle strength observed in this study. This is confirmed by the findings of previous investigations that have indicated that AGT is a safe method of remediating weight-bearing exercise [35]. Previous studies have examined the effect of AGT in the rehabilitation of young, healthy and physically active patients who have sustained acute musculoskeletal injuries [30,36]. Each of these studies utilized AGT as a way to successfully remediate or accelerate a progressive treatment program, with the level of AGT support, walking speed, and/or incline being adjusted as the patient progressed through their rehabilitation.

Our investigation is the first to examine the use of AGT support in the management of children with hemophilia, utilizing a standardized methodological approach (i.e., 5-minute warm-up, 20 minutes of treadmill walking, 0% incline).

This study was limited to 30 hemophilic children who were selected from the pediatrics out-patient clinic of the Hospital of Medical Rehabilitation and King Fahd Hospital, Almadinah Almonawara. Our sample was not large, the effect size was moderate to large (0.85, 0.83 and 0.553) for the knee extensors, flexors and PBBS respectively. According to the results of this study, further investigations and research studies are recommended: similar studies should be done on more sever types of hemophilia; longitudinal studies with a larger sample size are recommended; using more objective evaluative tools as Electromyography (EMG) during gait training on AGT to record muscular activity; other studies aiming to investigate the effect of AGT training on energy expenditure in these children are better to be applied.

5. CONCLUSIONS

On the basis of the present data, it is possible to conclude that gait training using AGT, in addition to a designed physical therapy program, is an effective therapeutic modality for improving muscle strength and functional balance in children with hemophilia. Although our sample was not large, the effect size was moderate to large (0.85 and 0.83) for the knee extensors and flexors respectively and 0.553 for PBBS.

COMPETING INTERESTS

Author has declared that no competing interests exist.

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