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INVESTIGATING THE EFFECTS OF STANDING TRAINING ON BODY FUNCTIONS AND ACTIVITY FOR NONAMBULATORY CHILDREN WITH MYELOMENINGOCELE

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ABSTRACT

Background: It was indicated in many studies that verticalization have positive effects such as preventing fractures, regulating cardiopulmonary functions, increasing the head control, and the facilitation of postural muscles in pediatric patients, however, no study showing the effect of supported standing in patients with myelomeningocele on body functions and activity was encountered. The aim of this study is to examine the effects of structured supported standing training in children with myelomeningocele on body functions and activity according to ICF-CY.

Method: Twenty-five children with MMC aged between 3 and 17, who were divided into two groups—SST and control. The supported standing training was given to supported standing group 2 hours a day for 8 weeks in addition to the routine physical therapy program. Body functions were assessed with the Trunk Impairment Scale, and activity levels were assessed with the Gross Motor Function Measurement-88 and Pediatric Functional Independence Measurement at the beginning of the study, at the end of 8 weeks and at the end of 12 weeks from beginning.

Results: The results of the structured supported standing training program during 8 weeks showed that children's body functions and activity increased statistically significantly in SST group ($p < 0.05$) but no difference was found between the two groups ($p > 0.05$).

Conclusion: The results show that supported standing training effects the body functions and activity positively. It is recommended to educate the families for the supported standing training to be added to the routine physiotherapy and rehabilitation program for children with MMC and continue the training at home.

Keywords: Myelomeningocele, Spina Bifida, Supported Standing, ICF-CY, Verticalization, Home-based training.

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INTRODUCTION

Myelomeningocele (MMC) is the most common encountered malformation among neural tube closing defects which cause structural anomalies at different levels in the spinal cord, cerebellum and brain stem [1, 2]. Motor, sensation, and cognitive problems are observed according to the lesion level [3]. With the developments in the medicine area nowadays, many children with MMC survive but 60% of them reach the adulthood period [3, 4]. While MMC is observed at the rate of 0.3-4 in 1000 live births around the world [5], the incidence of MMC in Turkey is 0,03% [6].

Due to the fact that nonambulatory children remain in sitting and lying positions for long periods of time, painful and costly complications develop in them [7]. While supported standing of nonambulatory children with MMC is discussed in some studies, some prefer wheelchair. Recent studies are on certain ensuring the verticalization of children. Standing programmes are used routinely as a part of postural management for children with disabled [8]. Paleg et al in 2013, Caulton et al in 2004, Pauly & Cremer in 2013 and Nordström et al in 2007 showed that supported standing has positive effects on body structure and functions such as an increase in psychological motivation, prevention of osteopenia and fractures, regulation of cardiopulmonary functions, increase in the visual perception, increase of the head control, facilitation of antigravity muscles and increase muscle strength and endurance and in addition verticalization could promote face-to face contact between people with and without disabled. [7-10]. Pauly & Cremer in 2013 stated the necessity of standing up as follows: "it seems to be obvious that verticalization is mandatory for mobilization and should be the goal of our therapeutic efforts." [9]. Nordström et al in 2007 emphasised that using a standing frame is optimal and promote an opportunity to change body position for people with limited mobility as different diagnosis [10].

The International Classification of Functioning children and youth version (ICF-CY) is a coding system developed by the World Health Organisation (WHO), which describes the health and function in children. The system which takes the biopsychosocial model as a basis involves body structure and functions, activity and participation, and environmental and personal factors [11, 12]. Nowadays, ICF-CY creates a structural basis for physiotherapy and rehabilitation assessments and programme planning. ICF-CY creates a conceptual framework to determine health information such as diagnosis and functionality and thus defines the problems of a child in relation to the function and anatomical characteristics, activity limitations and participation problems [11, 13]. Children with MMC have many problems on body structure and functions and activity limitations due to neuronal damage. Standing frames according to the ICF-CY are an environmental factor, it may facilitate or inhibit child's activity, participation, and body structures and functions [10]. Many activities depend on succeeding in standing and primarily weight transferring. The number of the studies in which children

with MMC are examined according to ICF-CY in the literature is very limited. For this reason, in the present study, it was aimed to investigate the effects of structured supported standing training on body functions and activity in children with MMC.

METHODS

Study design

This was a prospective study comparing structured supported standing training group with a control group. Participants were allocated randomly by using closed envelop method. A total of 25 children with myelomeningocele consisting of nonambulatory, according to the Hoffer criteria [14], children and families who agreed to participate in the study and signed the consent forms and applied to the Physiotherapy and Rehabilitation Department of Hacettepe University between April 2013 - April 2014 were assessed. The required approval was obtained from Hacettepe University Ethics Committee on Non-Interventional Clinic Research for the study to be performed (decision number: GO 13/75 - 06, 10.04.2013).

Participants

The criteria for the inclusion in the study were determined as: receiving myelomeningocele diagnosis, being at the age of 3-17, the absence of the joint deformity of the lower extremity which hinders the use of standing frame, being at a cognitive level that can ensure the understanding of the exercises, receiving the routine physiotherapy programme at a special training and rehabilitation centre 2 times a week (the disabled children have 2 times of week free physical therapy and rehabilitation section by government in Turkey) on a regular basis, having no secondary neurological diseases.

The criteria for the exclusion from the study were determined as: not following the routine physiotherapy program two days a week, not completing the 8-week training, not participating in the second and third evaluation and undergoing a surgery in the evaluation intervals.

Outcome measurements

The demographic information of the children was recorded. The children were assessed at the baseline (t1), after 8 weeks (t2) and after 12 weeks from baseline (t3) by the same physiotherapist.

The Trunk Impairment Scale (TIS); evaluates the static and dynamic balance and body coordination in the unsupported sitting position. An ordinal scale consisting of 2, 3 and 4 points is used for each item. The total score varies between 0 and 23. The total evaluation duration does not exceed 10 minutes [15, 16]. It was employed in the study to evaluate the body functions.

Gross Motor Function Measurement (GMFM-88); is a valid and reliable scale which is used to measure the changes in the gross motor function and which evaluates the body structure and functions, activity and participation sub headings according to ICF-CY. It consists of five sub-sections including a total of 88 items. A general scoring

system has been created that evaluates how much of each activity the child can complete. The scoring is with a four-point Likert scale. [17, 18]. GMFM-88 subheadings such as supine-prone-rolling (A), sitting (B), crawling (C) and the total were used for activity evaluation.

The Pediatric Functional Independence Measurement (WeeFIM); is a valid and reliable scale which evaluates the body structure and functions and mainly activity and participation function according to ICF-CY which measures the functional independence in self-care, mobility and cognition areas. Each activity is evaluated over 7 points. "7" indicates that the child is completely independent in daily living activities and "1" indicates that the child is completely dependent. [17, 19]. Self-care, mobility, and total scores were used for activity evaluation.

Intervention

The structured supported standing training programme, which was implemented to the intervention group (SST), consisted of play activities for strengthening (e.g.; unilateral and bilateral upper extremity activities such as playing basketball) and weight bearing (e. g.; reaching a toy from left side, ride side and anterior). The physiotherapist made the adjustments of the devices by going to the children's home and education was given to the child and his/her family in front of a mirror in order to provide alignment

during standing and protection. The program was taught to the family. This training was performed 2 hours each day of the week (3 times with 40-minute periods) as a home-based program during 8 weeks added routine physiotherapy. The continuity of the program was ensured with the weekly home visits and telephone calls.

Control group continued routine physiotherapy programme which consist of lower extremity joint range of motion exercises, trunk and upper extremity strengthening training and balance training twice a week.

Statistical Analysis

Statistical analyses were performed using the IBM SPSS for Windows Version 20.0 software program. Mann-Whitney U test was used for comparing the mean values of all the variables at baseline, eight weeks and twelve weeks between both groups. Friedman test was used for evaluating the effect of time in the groups. For the mean change over time, Mann-Whitney U-test was used for comparing groups at the time point. The significance level was accepted as $p < 0.05$.

RESULTS

A total of 25 children with MMC were included in this study. Three children excluded due to surgery (one child) and not coming second evaluation (two children) in the control group. The follow diagram was given in figure 1.

Figure 1. Flow Diagram of the children

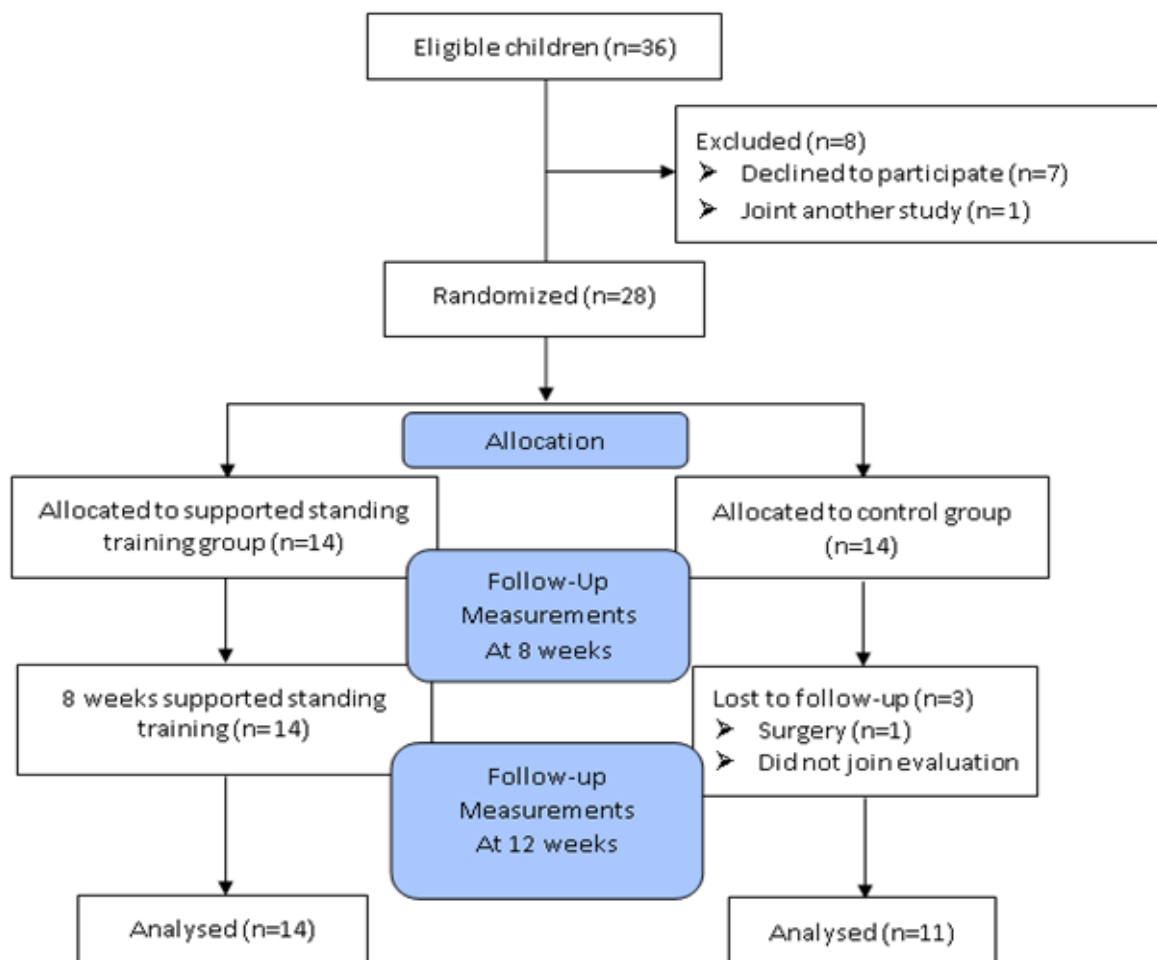


Table 1. Demographic characteristics of the children

	SST group (n=14)	Control group (n=11)	p-value
	Median (min-max)	Median (min-max)	
Age (year)	6 (3-15)	10 (3-15)	0.085
BMI (kg/m ²)	23 (19.8-26.6)	25 (18.1-28.3)	0.267

SST: Supported standing training, BMI: Body Mass Index

Demographic information

There was not a difference between the groups in terms of age and body mass index (p>0.05). Children’s demographic characteristics are shown in table 1. The median age of the children was 6 (min-max: 3-15) years and 10 (min-max: 3-15) years in SST group and control group respectively.

The groups are homogenous for lesion levels. The lesion levels are given by groups in table 2.

Table 2. Lesion levels of children

	SST group	Control group
Levels	n (%)	n (%)
Lumbosacral	5 (36)	2 (18)
Lumbal	8 (57)	7 (64)
Toracolumbal	1 (7)	2 (18)
Total	14 (100)	11 (100)

SST: Supported standing training

Body functions

A statistically significant improvement was found in the TIS static balance, dynamic balance and total scores in SST group (p<0.05). The post hoc tests showed that static balance (p=0.042), dynamic balance (p=0.038) and total score (p=0.014) increased between baseline and 8 weeks measurements. But there was no statistically significant difference between groups (p>0.05). The results are given in table 3.

Table 3. Comparison of change in outcomes at baseline, 8 weeks and 12 weeks for body functions

	Groups	Baseline	8 weeks	12 weeks	p-value
TIS		Median (min-max)	Median (min-max)	Median (min-max)	
TIS-static balance	SST group Control group p-value	5 (2-6) 5 (2-6) 1.000	5 (2-6) 5 (2-6) 0.727	5 (2-6) 5 (2-6) 0.647	0.022* 0.368
TIS-dynamic balance	SST group Control group p-value	0 (0-5) 3 (0-6) 0.58	1 (0-8) 3 (0-6) 0.222	1 (0-8) 3 (0-6) 0.244	0.003* 0.135
TIS-coordination	SST group Control group p-value	2 (0-4) 1 (0-2) 0.687	2 (0-4) 1 (0-2) 0.572	2 (0-4) 1 (0-2) 0.572	0.058 1.000
TIS-total	SST group Control group p-value	6 (2-12) 8 (2-14) 0.244	7 (2-15) 10 (2-14) 0.501	8 (2-15) 10 (2-14) 0.687	0.032* 0.135

SST: Supported standing training, TIS: Trunk Impairment Scale, *:p<0.05

Activity

The significant difference was found in terms of GMFM-A, GMFM-B and total scores in SST group. In the post hoc test results GMFM-A (p=0.047), GMFM-B (p=0.011) scores increased between baseline and 8 weeks measurements. The significant time effect was observed at GMFM total score (t1-t2, p=0.038; t1-t3, p=0.03). Also there was significant difference in WeeFIM self-care and total score (t1-t2, p=0.03; t1-t3, p=0.006) and total score (t1-t2, p=0.03; t1-t3, p=0.003) increased in the intervention group. But there was no significant difference between groups. The results are given in table 4.

Table 4. Comparison of change in outcomes baseline, 8 weeks and 12 weeks for activity

	Groups	Baseline	8 weeks	12 weeks	P-value
GMFM (%)		Median (min-max)	Median (min-max)	Median (min-max)	
GMFM-A	SST group Control group p-value	91.5 (73-99) 88.2 (52.9-96.1) 0.434	91.2 (74.5-100) 88.2 (52.9-96.1) 0.222	91.5 (73-99) 87 (52.9-95) 0.149	0.05* 0.779
GMFM-B	SST group Control group p-value	80.8 (61.7-100) 70 (41.7-98.3) 0.687	80.8 (62-100) 70 (41.7-98.3) 0.501	80.8 (62-100) 70 (41.7-97) 0.467	0.003* 0.607
GMFM-C	SST group Control group p-value	54.7 (2.4-94) 35.7 (7.1-71.4) 0.467	54.7 (2.4-95.2) 31 (7.1-71.4) 0.373	54.8 (2.4-94) 31 (7.1-71.4) 0.317	0.058 0.368
GMFM-T	SST group Control group p-value	42.5 (30-57.6) 36.6 (21-55.6) 0.403	43.4 (30-58) 36.4 (21-55.6) 0.317	43.5 (30-58) 36.4 (21-56.6) 0.244	0.032* 0.926
WeeFIM					
WeeFIM-self-care	SST group Control group p-value	31 (17-43) 35 (12-44) 0.291	31 (17-43) 36 (12-44) 0.344	31 (17-43) 36 (12-44) 0.373	0.001* 0.135
WeeFIM-mobility	SST group Control group p-value	19 (7-27) 23 (5-29) 0.893	19 (7-27) 23 (5-29) 0.893	19 (7-27) 23 (6-29) 0.893	0.223 0.368
Wee FIM-total	SST group Control group p-value	85 (62-105) 93 (53-108) 0.403	85 (62-105) 93 (53-108) 0.501	85 (62-105) 93 (53-108) 0.501	0.001* 0.097

SST: Supported standing training, GMFM: Gross Motor Function Measurement, WeeFIM: Pediatric Functional Independence Measurement, *:p<0.05

DISCUSSION

MMC is a problem which causes activity and participation limitations [20]. The present study shows that structured supported standing training program for children with MMC improves body function and reduces activity limitation.

In the present study, the SST was given in accordance with the literature as 2 hours a day for 8 weeks. Franki et al. in 2012 observed that when the standing and weight transferring training applied on SP children was examined varied between 2 weeks and 9 months. It was stated that an increase in body structure and functions, intestine activity with weight transferring training contributed to daily

functions and feeling good personally [21]. It was determined that standing program applied 3 times a day for 45 minutes to children with MMC was beneficial for preventing the lower extremity contractures and stimulating the bone development [22].

After the 8 weeks SST, the increase in trunk static balance, dynamic balance and total score according to TIS gives rise to the thought that trunk training with verticalization improves the trunk control and shows that the body function improves according to ICF-CY. In the systematic review of Ivanyi et al. in 2014, it was stated that supportive devices had a positive effect on the body structure and functions in children with spina bifida [23]. In Paleg et al. 2013 examined the effects of standing training in children with cerebral palsy according to ICF-CY. As a result, it was found out that standing for 30 minutes a day, 5 days a week regulated the muscular tonus of the lower extremities, improved the school performance, enabled the eye contact and increased the attention level. It was shown that standing for 40 minutes increased the functional blood circulation and decreased the respiratory distress, standing for 30-60 minutes regulated the intestinal functions, standing for 60 minutes ensured a positive contribution to providing the range of motion of the hip, knee and ankle joints, decreased the migration ratio by supporting the hip biomechanic structure and prevented subluxation, and along with these, standing for 60-90 minutes increased the bone mineral density and decreased osteoporosis [7]. Glickman et al. in 2010 showed that supported standing increased the bone mineral density, cardiopulmonary functions, bladder-intestinal functions, muscle strength and joint range of motion and decreased pressure ulcers [24].

The changes in GMFM A, B and total score after SST and WeeFIM self-care and total score shows that children transfer the function they have gained in the vertical position to activity. We consider that children perceive more in the vertical position and it is related to the fact that they try to ensure the trunk control during standing and use this control they have gained more effectively at the ground level. No activity and participation evaluation was encountered in the studies carried out on children with MMC [24]. In nonambulatory children with SP, it was shown that daily activities such as transferring, positioning, dressing, bathing, and going to the toilet could be performed more easily with the 6 weeks standing training applied for 6 days a week, 1 hour per day. In the study of Franki et al. in 2012, when the standing and weight transferring training applied on children with SP was examined according to ICF-CY, it was seen that performance and walking speed did not change in the activity and participation [21].

CONCLUSION

Severe motor loss and physical limitations in children with MMC affect body functions and activity such as ambulation and mobilisation, while it changes according to the lesion level in the medulla spinalis. Increasing the movement ability of the extremities and trunk with the motor loss is very important in terms of preventing the negative prob-

lems of especially weight bearing, standing, trunk training and mobilisation training in the physiotherapy and rehabilitation program in the early period. According to the results of our study, it is emphasised that SST has a positive effect on the body functions and activity. And it is recommended to educate the families for the supported standing training to be added to the routine physiotherapy and rehabilitation program for children with MMC and continue the training at home.

Limitations of the study

That the participation in the evaluations performed within the scope ICF-CY was not assessed, that the number of the patients who participated in the study was few caused the inability to demonstrate the results sufficiently. It is recommended to carry out research by increasing the number of participants and by adding the participation evaluation in future studies.

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