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Muscle Function of Lower Extremities in Children with Lumbar Spina Bifida: Impact on Functional Status

Lomber Spina Bifidalı Çocukların Alt Ekstremite Kas Fonksiyonları: Fonksiyonel Durum Üzerine Etkisi

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Abstract

Objective: The aim of this study was the evaluation of the impacts of muscle strength, muscle tone, and contractures of the lower extremities on ambulation and functional status in children with spina bifida (SB). The aims of this study were the determination of the demographical and clinical characteristics of Turkish children with spina bifida (SB) and the evaluation of the impacts of the clinical features on ambulation and functional status.
Material and Methods: Fifty-four children with SB were included in the study. The Functional Independence Measure for Children (WeeFIM), range of motion in lower extremities, and muscle tone and muscle strength of the lower extremities in all children and the ambulation level of children older than 36 months were evaluated.
Results: There were significant differences between the non-ambulant and functional ambulant children older than 36 months in terms of muscle strength and muscle tone ($p<0.001$). While no significant effects of muscle strength and muscle tone on WeeFIM scores were found ($p>0.05$), contracture was found to be effective on WeeFIM motor ($p=0.001$) and WeeFIM cognitive-total ($p<0.001$) scores by univariate analyses of variance. In children older than 36 months, significant effects of ambulatory status on WeeFIM cognitive ($p=0.019$) and WeeFIM motor-total ($p<0.001$) scores were found.
Conclusion: Muscle strength and muscle tone of lower extremities may be important for ambulation of children with SB. Ambulatory status and contractures may be important determinants for functional abilities of these children.
Key Words: Spina bifida, ambulation, disability

Özet

Amaç: Bu çalışmanın amacı spina bifidalı (SB) çocuklarda alt ekstremitelerin kas gücü, kas tonusu ve kontraktürlerinin ambulasyon ve fonksiyonel durum üzerine etkisinin değerlendirilmesi idi.
Gereç ve Yöntemler: Spina bifidalı 54 çocuk çalışmaya alındı. Pediatrik Fonksiyonel Bağımsızlık Ölçümü (PFBÖ), tüm çocuklarda alt ekstremitelerde eklem hareket açıklığı, alt ekstremitelerin kas tonusu ve kas gücü, 36 aydan büyük çocukların ambulasyon seviyeleri değerlendirildi.
Bulgular: Ambule olmayan ve fonksiyonel ambule olan 36 aydan büyük çocuklar arasında kas gücü ve kas tonusu açısından anlamlı fark vardı ($p<0,001$). Kas gücü ve kas tonusunun PFBÖ skorları üzerine anlamlı etkisi bulunmazken, kontraktür PFBÖ motor ($p=0,001$) ve PFBÖ kognitif-toplam ($p<0,001$) skorları üzerine etkisi bulundu. Otuz altı aydan büyük olan çocuklarda, ambulasyonun PFBÖ kognitif ($p=0,019$) ve PFBÖ motor-toplam ($p<0,001$) skorları üzerine anlamlı etkisinin olduğu bulundu.
Sonuç: Alt ekstremitelerin kas gücü ve kas tonusu, SB'li çocukların ambulasyonunda önemli olabilir. Ambulasyon ve kontraktürler, bu çocukların fonksiyonel yeteneklerinin önemli belirleyicileri olabilirler.
Anahtar Kelimeler: Spina bifida, ambulasyon, dizabilite

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Introduction

Spina bifida (SB) is a major birth defect that occurs when the spinal cord does not close properly during embryonic development (1). It is a complex congenital disorder that represents a broad spectrum of neural tube defects, including SB aperta and SB occulta. SB aperta is a midline defect that communicates with the external environment and includes myelomeningocele and meningocele (2). The prevalence estimate of SB is 3.5 per 10,000 live births in the USA (1), and it has been reported that in Turkey, the neural tube defect prevalence is 3-9 in 1,000 live births (3-6).

Children with SB have a range of disabilities, depending on where in the spinal column formation the defect is located and whether it is closed or open (7). Children with SB do not present simply with flaccid paraplegia below their anatomic lesion. Only one-third presents with flaccid paralysis, while most have a combination of upper and lower motor neuron signs. Some have voluntary motor control below other segments of paralysis and sensory loss. The level of neurologic impairment influences medical providers' expectations for functional outcome, as well as musculoskeletal deformities and complications to anticipate (2). Infants and children with SB have a high risk of lifelong cognitive and physical disabilities, and only a minority of these children is able to function independently as adults (1,8).

The majority of children with SB has lumbosacral level vertebral lesions, and approximately one-fourth of patients present with lumbar-level involvement (2). These patients may exhibit a variety of motor abnormalities, including incomplete flaccid paraplegia, spasticity, and mixed paraplegia and spasticity, with asymmetrical involvement on either side (9). It was reported that patients with third-lumbar and higher levels of paraplegia usually become non-ambulatory, whereas patients with fourth-lumbar or lower levels of paraplegia usually remain functional walkers (10).

Spina bifida is a non-progressive, permanent neurodevelopmental disability, and it causes associated impairments involving several domains of body functions (11). These impairments cause a restriction in the performance of daily activities, such as standing, ambulation, and the voluntary control of bladder and bowel functions (12). Accompanying cognitive disorders also lead to increased dependency on additional care. Although the prevalence rate of neural tube defects is very high for Turkey (6), there are limited studies in the literature that have evaluated clinical features of children with neural tube defects and the impacts of these features on functional status in Turkey. Previously we studied the impact of functional disability of Turkish children with SB on parents'psychological status and family functioning but the relation between the clinical features and functional status of these children was not investigated (13). The aim of this study was to determine the effects of muscle strength, muscle tone, and contractures of the lower extremities on ambulation and functional status in Turkish children with SB.

Material and Methods

The study was conducted at the outpatient clinic at the Department of Physical Medicine and Rehabilitation of Medical Faculty of Ondokuz Mayıs University between January 2011 and June 2012. The Ondokuz Mayis University Medical Research Ethics Committee approved the study protocol, and written informed consent was obtained from the primary caregivers of the children. The participants were 54 children with SB, and children with SB occulta were excluded from the study. Data were collected on age, sex, body mass index (BMI), number of siblings (disabled/non-disabled), operations, and lesion level of the children. All parents were questioned about age, working status, and educational level, and they were also asked to be informed about whether the diagnosis of SB was made prenatally.

The child's version of the adult Functional Independence Measure (WeeFIM) is a standardized assessment of pediatric functional independence, designed to evaluate children with special healthcare needs, primarily those diagnosed with acquired or congenital deficits or developmental delays. It includes 18 items covering six areas: self-care (eating, grooming, bathing, dressing upper body, dressing lower body, toileting); sphincter control (bladder management, bowel management); transfer (chair/bed/wheelchair transfer, toilet transfer, tub/shower transfer); locomotion (crawling/walking/wheelchair, stair climbing); communication (comprehension, expression); and social cognition (social interaction, problem solving, memory). A 7-level ordinal rating system ranging from 1 (total assistance) to 7 (complete independence) is used to score the performance on each item. The Wee-FIM consists of two dimensions: motor and cognitive. The motor scale includes self-care, sphincter control, transfer, and locomotion items; the cognitive scale includes communication and social cognition items. It was found as a reliable and valid instrument in Turkish non-disabled children and children with cerebral palsy (14-16).

Range of motion in lower extremities was measured in a standardized way with a two-legged 360-degree goniometer. Loss of range of motion of 5% or more per joint in one or both extremities was defined as a contracture (17).

Muscle tone of lower extremities was scored on a 5-point scale ranging from severe hypotonia (1), mild hypotonia (2), normal tone (3), and mild hypertonia (4) to severe hypertonia (5). Both mild or severe hypotonia and hypertonia were classified as an abnormal score (17).

Muscle strength of lower extremities was graded as 0-5, according to the standard Manual Muscle Testing, as described by Hislop and Montgomery (18). In children under the age of 5 years, muscle grading on a 6-point scale is not reliable. Therefore, in these patients, strength was graded as absent, weak, or full strength, as advocated by McDonald et al. (19). In the lower extremities, flexors, extensors, and abductors of the hip, as well as extensors of the knee and dorsal flexors of the ankle joint, were tested. Knee extensor muscles were used as indicator muscles for lower extremity muscle strength (17). Children with full strength or grade 5 were classified as having normal muscle strength (17).

Ambulation level of children older than 36 months was scored according to the criteria of Hoffer et al. (20). Patients were grouped into non-ambulators (completely wheelchair-dependent or walking in therapeutic situations) and functional ambulators (household or community walkers) (17).

Statistical Analysis

Statistical analyses were performed with Statistical Package for the Social Sciences (SPSS) 16.0 for windows. Descriptive data were presented as mean±standard deviation (SD) or median (minimum-maximum). The Kolmogorov-Smirnov test was used to analyze the normal distribution assumption of quantitative outcomes, and all data were not normally distributed. Chi-square test was used to compare the clinical features of children older than 36 months. Univariate analyses of variance were used to assess the effects of muscle strength, muscle tone, contracture, and ambulatory status on WeeFIM scores. In order to have a statistical power of 0.99 and $p<0.05$, it was calculated that 50 children in the study were required to detect the disability level of the participants. P values less than 0.05 were considered statistically significant.

Results

The 54 children with SB (30 females, 24 males) had a mean age of 52.2 months (SD 43.31, range 7-144 months, median 39 months). The demographic and clinical characteristics of the children are shown in Table 1. The percentages of children with normal lower extremity muscle strength and normal lower extremity muscle tone were 27.8% and 51.9%, respectively. Additionally, 63% of the children had no contractures in the lower extremities. The lesion level of all patients was lumbar, and all patients underwent primary closure and shunt surgery within 48 hours after birth. Among the parents, 50% reported that they had been informed during pregnancy that their fetus had SB, and there was no difference between fathers and mothers who had this information. Of the families, 21 (38.9%) had no more children, and 33 (61.1%) patients with SB had further siblings. In all cases, the siblings were healthy but one. Of the children older than 36 months (n=30), 11 (36.7%) were ambulant and 19 (63.3%) were non-ambulant. There were significant differences between ambulant and non-ambulant children in terms of muscle strength ($p<0.001$) and muscle tone ($p<0.001$). The presence of contracture was not different in ambulant and non-ambulant children ($p>0.05$). Table 2 presents the comparison of the clinical features of the non-ambulant and functional ambulant children older than 36 months.

By univariate analyses of variance, no significant effects of muscle strength and muscle tone on WeeFIM scores were found ($p>0.05$). Contracture was found to be effective on WeeFIM motor ($p=0.001$), WeeFIM cognitive ($p=0.003$), and WeeFIM total ($p<0.001$) scores by univariate analyses of variance. In children older than 36 months, a significant effect of ambulatory status on WeeFIM motor ($p<0.001$), WeeFIM cognitive ($p=0.019$), and WeeFIM total ($p<0.001$) scores was found by univariate analyses of variance (Table 3).

Discussion

Neural tube defects are the second most common group of serious birth defects, and SB is one of the most common of them (8). Some studies have been published on the prevalence, incidence, and risk factors of neural tube defects in our country (3,4,6,21). This study explored the muscle functions of lower

Table 1. Demographic and clinical characteristics of the patients and parents

Characteristics	n=54	
Age (months)		
Mean±SD	52.5±43.31	
Median (min-max)	39 (7-144)	
	n	**%**
Muscle strength		
Normal	15	27.8
Abnormal	39	72.2
Muscle tone		
Normal	28	51.9
Abnormal	26	48.1
Contracture		
Yes	20	37.0
No	34	63.0
Ambulatory status in children≥36 months		
Non-ambulant	19	63.3
Functional ambulant	11	36.7
	Mean±SD	**Median (min-max)**
WeeFIM		
Motor	32.65±23.36	23.5 (10-96)
Cognitive	23.61±10.68	27 (5-35)
Total	56.26±32.26	48 (18-131)
Parents' age (year)		
Mothers	29.6±5.06	29 (21-42)
Fathers	33.8±5.75	33 (23-49)
	n	**%**
Parents' occupations		
Mothers (n=54)		
Housewife	54	100
Fathers (n=54)		
Government official	30	55.5
Other	24	44.5
Parents' educations		
Mothers (n=54)		
Primary education	40	74.1
Secondary education	9	16.7
College	5	9.2
Fathers (n=54)		
Literate	1	1.9
Primary education	26	48.1
Secondary education	15	27.8
College	12	22.2

Mean±SD: mean±standard deviation, median (min-max): median (minimum-maximum);
p value is significant when <0.05

Table 2. Comparison of the clinical features of children older than 36 months

Characteristics	Children older than 36 months (n=30)		p
	Ambulant (n=11)	Non-ambulant (n=19)	
	n (%)		
Muscle strength			
Normal	7 (63.6)	0	<0.001
Abnormal	4 (36.4)	19 (100)	
Muscle tone			
Normal	11 (100)	3 (15.8)	<0.001
Abnormal	0	16 (84.2)	
Contracture			
Yes	6 (54.5)	5 (26.3)	0.125
No	5 (45.5)	14 (73.7)	

p value is significant when <0.05

Table 3. Univariate p values of clinical features for WeeFIM scores

Predictors	Univariate p value		
	WeeFIM motor	WeeFIM cognitive	WeeFIM total
Muscle strength	0.060	0.591	0.101
Muscle tone	0.613	0.206	0.382
Contracture	0.001	0.003	<0.001
Ambulatory status in children ≥36 months	<0.001	0.019	<0.001

extremities affecting ambulation and functional status in Turkish children with SB.

In a study by Bulbul et al. (21), it was found that meningomyelocele was mostly present in the lumbar area, which is similar to previous studies. In the current trial, all patients had lumbar lesions, and neurological and physical examinations of the upper extremities and trunk were normal. It was shown that the prevalence of neural tube defects is high in families with low educational level in our country (3,4,6,21). In concordance with these studies, 74.1% of mothers and 48.1% of fathers had primary education in our study. Most cases of anencephaly and spina bifida can be detected through prenatal screening methods, such as fetal ultrasound scanning (8). In this study, 50% of parents had information that they will have children with SB. Bulbul et al. (21) reported that 20 of 28 neonates had been diagnosed with meningomyelocele during the antenatal period, and none of the families terminated the pregnancy in their study. Similarly, in our study, parents reported that they did not want to terminate the pregnancy, which can be attributed to the socio-cultural characteristics of our country, as suggested by Bulbul et al. (21).

In the current study, functional outcome of the children with SB was assessed by WeeFIM, which measures functional abilities and limitations when performing activities of daily living, taking into account caregiver assistance and the use of special equipment (14). It was shown that mental status, contractures, and muscle strength of lower extremities were the most important determinants for independence in self-care and mobility (17,22). Schoenmakers et al. (17) found that knee extensors were associated with independence in mobility. Contrarily, in the current trial, muscle strength and muscle tone had no effect on WeeFIM scores. On the other hand, contracture was found to be a predictor variable of functional disability level of children with SB in this study. This finding is in line with previous studies suggesting that contractures are an important determinant for functional abilities (17,23,24). It can be suggested that prevention of contractures with surgical procedures and/or physiotherapy programs may be important for the functional improvement of children with SB. It was reported that motor function appeared to be important for complete independence in self-care (17). Although muscle strength and muscle tone were not found to be predictor variables of functional disability level in this study, physiotherapy programs should also be designed to increase the power of the lower extremity muscles.

It was reported that maximizing ambulatory function will promote future independent living and higher QOL in children with myelomeningocele (22). Tsai et al. (9) found the relation between PEDI scores and walking ability. Similarly, we found that ambulatory status had a significant effect on functional disability level of children with SB older than 36 months. It can be concluded that better ambulatory function may lead to improved management of activities of daily living.

It is well known that the level of the neurological lesion and the strength of lower muscles are the most important factors influencing ambulatory status (17). It was also reported that spasticity might be a contributing factor to contractures, which are associated with loss of walking ability. In the current trial, non-ambulant children with SB older than 36 years had poor muscle strength and abnormal muscle tone compared to those who were ambulant. Contrarily, contracture was not to be found different in ambulant and non-ambulant children older than 36 months. Since the lesion levels of all children were lumbar and were not classified as upper or lower lumbar, it is not possible to comment about the effect of lesion level on ambulatory status in this study. Previous studies already showed that lesion level had less importance than muscle strength of lower extremities with regard to independence in mobility, and children with a similar level of motor paresis did not always develop similar ambulatory levels (17,25).

The major limitation of this study is that the lesion level of all children was lumbar; thus, the results can not be generalized to the general SB population. Children with SB have problems related to hydrocephalus, neurogenic bladder, kidney involvement, orthopedic complications, and psychosocial consequences (8). Another limitation of this study is that these complications were not evaluated. The most powerful aspect of our study is that it is one of the few studies that have evaluated some clinical and demographic features of children with SB in our country. In this study, homogeneous participants, which were children with lumbar SB, were evaluated. Many studies on SB are based

solely on maternal reports; in this study, the data of the fathers were also assessed.

Based on the results of the current study, muscle strength and muscle tone of lower extremities are important for ambulation of children with SB. Ambulatory status and contractures were important determinants for functional abilities of these children. Prevention of contractures and improving muscle strength and muscle tone should be considered in the treatment of children with SB. Better ambulation appeared to contribute to more functional independence, and it should be kept in mind when planning physiotherapy, surgical procedures, or medication in children with SB.

Ethics Committee Approval: Ethics committee approval was received for this study from the ethics committee of Ondokuz Mayıs University Medical Research Ethics Committee.

Informed Consent: Written informed consent was obtained from the primary caregivers of the children who participated in this study.

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