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EVALUATION OF MUSCLE STRENGTH IN PATIENTS WITH SPINAL MUSCULAR ATROPHY

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The first aim of this study was to evaluate the muscular strength in order to document the strength scores for patients with spinal muscular atrophy (SMA). The second aim was to analyze the differences in average strength score among the musculatures at various body parts. Twenty-five patients with type Ⅱ SMA (mean age of 13.7 ± 7.0 years) and 15 patients with type Ⅲ SMA (mean age of 13.7± 8.2 years) were enrolled to undergo a muscle strength evaluation using the method of manual muscle testing. For each patient, the strength of 71 muscle groups were graded using a numerical ordinal scale to obtain individual muscle group scores, average scores of the arms, legs, the musculatures around the limbs' joints, the arm flexors and extensors, and leg flexors and extensors. The data were analyzed using a statistic model of multivariate analysis of variance and paired t-test. All data were administrated respectively for type Ⅱ and type Ⅲ SMA. The results showed that in both types of patients, the trunk muscles and the musculatures around the hips possessed lower strength scores, whereas elbow flexors, wrist flexors and extensors, finger flexors, and diaphragms exhibited relatively higher strength scores. In type Ⅱ SMA the average scores of the musculatures of the left side arms was lower than those of the right side at the joints of scapulas, elbows and wrists. Our data also showed the arm flexor scores were higher than the arm extensor scores in both types, whereas significant higher score of the leg flexor in comparison to the leg extensor was only found in the patients with type Ⅱ SMA. The profiles of muscle strength and strength differences over various body parts in the patients with type Ⅱ SMA as well as the type Ⅲ SMA were obtained from this study.

Key words: spinal muscular atrophy, muscle strength

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Spinal muscular atrophy (SMA) is an autosomal recessive neuromuscular disease due to degeneration of anterior horn cells in the spinal cord [1]. It has been recognized as being one of the most common autosomal neuromuscular disorders causing physical disabilities among children [2]. SMA can be classified into three main clinical types on the basis of age at onset and the best motor function achievement [2,3]. Patients with type I (severe type) SMA are never able to sit and generally die before two years of age. In the patients with the other two types, which are the type Ⅱ (milder type) SMA (patients who are able to sit but are never unable to walk without assistance) and type Ⅲ (mildest type) SMA (patients who are able to walk without help), the course of this disease tends to be slower [2,4].

Muscular weakness and deterioration of physical function are the obvious clinical features found in patients with SMA [1,2]. To date, there still exists no definitive medical management that is able to treat the muscle weakness. Clinically, supportive therapy to maximize and maintain the patients' present physical ability is strongly recommended by clinicians [5,

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6]. Understanding the profile of muscle strength in these patients is critically important in many ways, such as defining the patients' systemic physical capability and reflecting the change in strength if a specific clinical treatment is given. Although muscle testing for understanding the strength loss among patients with neuromuscular diseases is available in many literatures [7-10], there is still a lack of research on systemic muscle testing for comprehending the characteristic of muscle weakness distribution over various body parts in patients with SMA. The first aim of this study was to evaluate the muscle strength in order to document the strength scores in patients with spinal muscular atrophy (SMA). The second aim was to analyze the differences in average strength score among the musculatures at various body parts.

MATERIALS AND METHODS

Subjects and their characteristics of physical functions

The patients were diagnosed by a pediatric neurologist in an outpatient clinic and were enrolled in this study. All of them exhibited a homozygous deletion of the survival motor neuron gene, which is a critical criterion for diagnosis of SMA [9]. All the patients were over 4 years old because of the inability of younger children to cooperate with the strength evaluation process. There were 13 males and 12 females meeting the diagnosis of type Ⅱ SMA with a mean age of 13.7 ± 7.0 years. Nine males and 6 females were diagnosed as type Ⅲ SMA with a mean age of 13.7 ± 8.2 years.

The patients presented a range of physical abilities from independent walking to totally dependency in daily living activities. For the 25 patients with type Ⅱ SMA, 8 of them were able to raise their arms above their head. Eight patients had the best ability of raise their hands to their mouth and 9 of them could only use their hands to manipulate small things. In this type of patients, 24 were restricted to wheelchairs, whereas only one patient could stand with a long leg brace. Regarding the 15 patients with type Ⅲ SMA, 14 were able to raise arms above head, whereas one patient could only to raise hands to mouth. Fourteen patients with type Ⅲ SMA had the ability of walk with or without assistance, while the remaining one was restricted to a wheelchair.

Measurement of muscle strength

Patients underwent manual muscle testing (MMT) to evaluate the strength of muscle groups at necks and trunks and also the musculatures around the limbs' joints, which were the scapulas, shoulders, elbows, wrists, fingers, hips, knees, ankles and toes. The MMT methodology has become a clinical standard for measuring muscular strength [11,12] and is especially useful for documenting the extent of strength loss in weak muscles.

For the strength measurement, the patients were prepared in a separated and quiet room. The procedures for conducting the measurement were standardized using the system published by the Hislop *et al.* [13] The patients' testing positions were organized in the order of supine position, side-lying position, prone position, sitting position and standing position, so that positional changes in a test sequence were minimized. Prior to the measurement, the actions of the measured muscle groups were illustrated and demonstrated to the patients individually in order to make sure they could perform the tests correctly. Afterward, the patients performed each tested motion two or three times and the best performance was chosen for recording. A total of 71 muscle groups were measured. During the test sequence, there was a few minutes break for the patients to rest between different positions. A total of 30 to 40 minutes would be taken to complete a test sequence.

The measured muscle groups were graded using a numerical ordinal scale that consisted of 12 graded scores [14] ranging from zero, which represented no muscle contraction activity, to five, which represented a normal response to the test (Table 1). The grading is based on a system in which the ability of a measured muscle groups to hold the tested part in a given position against gravity establishes a score of 3. For the grades above this score, additional manual resistance is applied to the tested part, whereas muscles that were weaker than the score of 3 would be prepared for involving motions in the horizontal plane in which the resistance by gravity was eliminated. The scores of individual muscle groups could be added to get summations of score. Average score of a given body part was the product of the summation of individual muscle group scores divided by the number of added individual scores.

A physical therapist performed all the strength measurements in order to maximize the reliability and consistency of the evaluation procedure. The reliability of manual muscle testing for all the measured muscles was established by calculating the intra-class correlation coefficient (ICC 3,1) [15] on five patients with SMA prior to the start of the study. The mea-

surements demonstrated moderate to good intra-rater reliability with the ICC's ranging from 0.70 to 0.99.

Statistics

Since the clinical features of physical abilities in patients with type Ⅱ and type Ⅲ SMA are dissimilar [2,3], all data in this study were administrated respectively for the two types of SMA in order to understand their respective profiles of muscle strength. The second author, a statistician, of this study, analyzed the data of strength measurement. A model of multivariate analysis of variance (MANOVA) [16] was used for simultaneously comparing the average strength scores of the musculatures between the right and left side joints of scapulas, shoulders, elbows, wrists and fingers. This statistic model was also administrated to compare average scores of the musculatures between the two side joints of hips, knees, ankles and toes. In addition, the average score differences between arm flexors and extensors, and also between leg flexors and extensors were analyzed using paired t-test.

The statistical significance of the Bonferroni adjustment [17] was determined using an alpha level of 0.01 for all the data analyses.

RESULTS

The 71 individual muscle group scores by type of SMA are shown in Table 2. In type Ⅱ SMA, the individual muscle group scores of the trunk muscles and the muscle groups around the hips and knees were as low as about one except the hip adductors and knee flexors, which possessed relative higher scores among the hip and knee muscle groups. The muscle groups that had higher individual scores in type Ⅱ SMA were found as the diaphragms, elbow flexors, and the musculatures around wrists and fingers; such scores ranged from 2.5 to 3.2.

For type Ⅲ SMA, hip flexors had the lowest individual score of 1.9. Individual scores of the various trunk muscles and the musculatures around the hips were also relatively lower, whereas diaphragms, neck extensors, ankle dorsiflexors and the musculatures around elbows, wrists and fingers possessed higher scores among the measured muscle groups.

Table 3 displays the average strength scores of the musculatures around the joints at the right and left sides of arms and legs. For type Ⅱ SMA, the average strength score of the left arm was significantly lower than that of the right arm. Significant differences in the average strength score were also found between the musculatures at the two side scapulas, elbows and wrists (Table 3). In type Ⅲ SMA, no significant score differences were found between the right and left joints of the arms, even though a statistical tendency to score difference between the two shoulders existed ($p=0.018$, not shown in the Table). For both types of SMA, no significant

Table 1. Strength grading for muscle groups

Scores	Degree of strength
5.00	Normal strength
4.50	Barely detectable weakness
4.00	Muscle is weak but moves the joint against gravity and resistance
3.66	Same as 4 but weaker
3.33	Muscle moves the joint against gravity and an additional small amount of resistance but collapses abruptly.
3.00	Muscle moves the joint fully against gravity but cannot move against resistance
2.66	Muscle partially moves the joint against gravity
2.33	When gravity is eliminated, muscle moves the joint fully and an additional resistance
2.00	Muscle moves the joint fully when gravity is eliminated
1.50	Muscle moves a partial range when gravity is eliminated
1.00	A flicker of movement is seen or felt in the muscle
0.00	No movement or muscle contracture

(The Table was established according to the reference no. 14)

Table 2. The individual muscle group scores in type Ⅱ and type Ⅲ SMA

Muscle groups	Type Ⅱ SMA (n=25)		Type Ⅲ SMA (n=15)	
	Right	Left	Right	Left
Neck extensor	1.9 ± 0.7		3.4 ± 0.8	
Neck flexor	1.9 ± 0.6		3.1 ± 1.1	
Neck rotator	1.8 ± 0.6		3.2 ± 0.9	
Trunk extensor	0.8 ± 0.5		2.6 ± 1.4	
Trunk flexor	0.9 ± 0.3		2.4 ± 1.3	
Trunk rotator	0.9 ± 0.4		2.4 ± 1.3	
Diaphragm	3.2 ± 0.6		3.7 ± 0.8	
Scapular abductor	1.7 ± 0.7	1.5 ± 0.6	2.5 ± 1.2	2.2± 1.3
Scapular adductor	1.8 ± 0.7	1.7 ± 0.7	2.4 ± 1.4	2.4± 1.4
Shoulder flexor	1.8 ± 0.9	1.6 ± 0.9	3.2 ± 0.7	3.1± 1.0
Shoulder extensor	1.4 ± 0.7	1.5 ± 0.7	2.7 ± 1.0	2.6± 1.2
Shoulder abductor	1.7 ± 0.8	1.6 ± 0.8	3.2 ± 0.7	3.2± 0.8
Shoulder adductor	1.7 ± 0.7	1.7 ± 0.6	3.0 ± 1.0	2.9± 1.2
Shoulder external rotator	2.3 ± 1.1	2.2 ± 1.2	3.2 ± 0.9	3.0± 1.0
Shoulder internal rotator	2.1 ± 1.0	2.0 ± 1.1	2.9 ± 1.0	2.5± 1.1
Elbow flexor	3.1 ± 0.5	2.8 ± 0.6	3.4 ± 0.6	3.3± 0.6
Elbow extensor	1.8 ± 0.9	1.6 ± 0.9	3.1 ± 0.9	3.0± 1.0
Elbow supinator	2.6 ± 0.8	2.6 ± 0.9	3.4 ± 0.6	3.3± 0.7
Elbow pronator	2.6 ± 1.0	2.6 ± 1.0	3.5 ± 0.6	3.4± 0.7
Wrist flexor	3.0 ± 0.5	2.9 ± 0.6	3.5 ± 0.6	3.5± 0.6
Wrist extensor	2.8 ± 0.7	2.6 ± 0.9	3.5 ± 0.6	3.5± 0.6
Finger flexor	2.8 ± 0.7	2.7 ± 0.7	3.6 ± 0.5	3.5± 0.5
Finger extensor	2.5 ± 0.7	2.5 ± 0.8	3.2 ± 0.6	3.2± 0.5
Thumb flexor	2.5 ± 0.9	2.5 ± 0.8	3.2 ± 0.6	3.2± 0.6
Thumb extensor	2.5 ± 0.9	2.5 ± 0.8	3.1 ± 0.6	3.1± 0.6
Thumb abductor	2.5 ± 0.1	2.5 ± 1.0	3.1 ± 0.7	3.1± 0.7
Thumb adductor	2.5 ± 1.0	2.5 ± 1.0	3.1 ± 0.7	3.1± 0.7
Hip flexor	1.2 ± 0.5	1.3 ± 0.5	1.9 ± 0.8	1.8± 0.9
Hip extensor	1.1 ± 0.6	1.1 ± 0.5	2.6 ± 1.1	2.7± 1.2
Hip abductor	1.1 ± 0.6	1.1 ± 0.6	2.6 ± 0.9	2.6± 0.9
Hip adductor	2.0 ± 0.8	2.0 ± 0.4	2.3 ± 0.9	2.3± 0.8
Hip external rotator	1.0 ± 0.8	1.1 ± 0.6	2.2 ± 0.9	2.2± 0.9
Hip internal rotator	1.1 ± 0.8	1.2 ± 0.6	2.3 ± 0.8	2.2± 0.8
Knee flexor	2.1 ± 0.9	2.0 ± 1.0	3.1 ± 0.8	2.9± 0.8
Knee extensor	0.9 ± 0.7	0.8 ± 0.7	2.7 ± 1.1	2.6± 1.1
Ankle dorsiflexor	1.9 ± 1.1	1.9 ± 1.1	3.4 ± 0.5	3.3± 0.6
Ankle plantar flexor	2.1 ± 0.3	2.0 ± 0.4	2.7 ± 0.7	2.7± 0.7
Toe flexor	2.2 ± 0.9	2.2 ± 0.8	3.3 ± 0.8	3.3± 0.7
Toe extensor	1.9 ± 1.0	1.7 ± 1.1	3.3 ± 0.9	3.3± 0.8

Table 3.The average strength scores of the musculatures around the joints at the right and left sides of arms and legs in type Ⅱ and type Ⅲ SMA

Body parts	Type Ⅱ SMA (n=25)		Type Ⅲ SMA (n=15)	
	Right	Left	Right	Left
Arm				
Whole arm	2.4 ± 0.7	2.1 ± 0.6 *	3.2 ± 0.6	3.1 ± 0.7
Scapula	1.9 ± 0.8	1.7 ± 0.7 *	2.8 ± 1.0	2.7 ± 1.1
Shoulder	1.8 ± 0.7	1.7 ± 0.8	3.0 ± 0.8	2.8 ± 0.9
Elbow	2.6 ± 0.7	2.4 ± 0.7 *	3.4 ± 0.6	3.3 ± 0.7
Wrist	3.0 ± 0.5	2.7 ± 0.7 *	3.5 ± 0.6	3.5 ± 0.6
Finger	2.5 ± 0.7	2.3 ± 0.8	3.3 ± 0.6	3.2 ± 0.5
Leg				
Whole leg	1.7 ± 0.6	1.6 ± 0.6	2.9 ± 0.7	2.8 ± 0.7
Hip	1.2 ± 0.5	1.2 ± 0.4	2.3 ± 0.7	2.3 ± 0.7
Knee	1.5 ± 0.7	1.3 ± 0.7	2.9 ± 0.8	2.8 ± 0.8
Ankle	1.9 ± 0.7	1.8 ± 0.7	3.1 ± 0.7	3.1 ± 0.7
Toe	2.1 ± 0.8	2.0 ± 0.7	3.3 ± 0.8	3.2 ± 0.6

* p value<0.01, multivariate analysis of variance for comparing the average strength scores for the right and left sides

Table 4. The average strength scores of the flexors and extensors at the arms and legs in type Ⅱ and type Ⅲ SMA

Body parts	Type Ⅱ SMA (n=25)		Type Ⅲ SMA (n=15)	
	Flexor	Extensor	Flexor	Extensor
Arm	2.4 ± 0.7	2.0 ± 0.7 *	3.3 ± 0.6	3.0 ± 0.8 *
Leg	1.5 ± 0.6	0.3 ± 0.5*	2.6 ± 0.6	2.5 ± 0.8

* p value<0.01, paired t-test for the mutual antagonists of flexors and extensors

differences in average scores were found between the musculatures at the right and left joints of the legs.

For both types of SMA, arm flexor average scores were higher than the arm extensor average scores, whereas significant higher average score of the leg flexor in comparison to the leg extensor was only found in the patients with type Ⅱ SMA (Table 4).

DISCUSSION

This study delineated the respective systemic muscle strength in patients with type Ⅱ and type Ⅲ SMA. The strength scores of the measured muscle groups showed marked individual variations. The scores, which were less than 5 revealed such muscle groups had various extents of muscle weakness. In our patients, the trunk and hip muscles were the weakest muscle groups among the measured muscles. Of the other muscle groups, the elbow flexors, wrist flexors and extensors, finger flexors, and diaphragms exhibited less strength loss. In type Ⅱ SMA, hip adductors were relatively stronger compared with the other hip muscles. Liu *et al.* [18] reported on magnetic resonance imaging findings for type Ⅱ SMA in the lower extremity, which study showed severe atrophy of the thigh and calf muscle bundles with selective preservation of the adductor longus muscle, as well as the strongest hip adductors among the hip muscles.

The comparison of the right side muscle strength to the left side has not been reported in other similar researches. The significant average score difference between the two sides of arms was noted in our pa-

tients with type Ⅱ SMA. This result revealed muscle weakness over bilateral arms was asymmetric in these patients. Such strength differences occurred between the musculatures at the bilateral scapulas, elbows and wrists, which were the relative stronger body parts and may be frequently used for involving physical activities in these patients. Whether the strength asymmetry is just one of the natural features in SMA, or if prominent use of unilateral arm would lead to asymmetric muscle strength in this type of patients, data concerning such issues did not present in this study. Factors that contribute to the existence of bilateral asymmetry of muscle weakness are worth further study.

The comparison of muscle strength between flexors and extensors also has not been reported in previous studies. The significant difference in average strength scores between the arms and legs flexors and extensors found in this study revealed the imbalance of muscle strength existed between these mutual antagonists. Such strength imbalance between joint antagonists may be easily prone to develop joint contracture, which is the common and severe problem found in the patients with neuromuscular diseases [19]. With regard to the leg flexors and extensors in the patients with type Ⅲ SMA, the preservation of the walking ability in these patients may be a significant factor associating to the finding of the non-significant strength difference between these mutual antagonists. Since walking movements could facilitate and maintain the extensor strength of legs which may reduce the strength difference to the stronger leg flexors. Studies of the exact relationships between the imbalance strength of mutual antagonists and the existence of joint contracture as well as the disability of physical function should be required in further researches. Other future studies would be addressed to obtain additional data for understanding the changes of muscle strength in patients with SMA.

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脊髓肌肉萎縮症患者的肌力評估

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本研究的目的爲評估脊髓肌肉萎縮症（簡稱SMA）患者的肌力以取得肌力分數，並分析身體各部位平均肌力分數的差異情況。研究對象爲25名第Ⅱ型SMA（平均年齡爲13.7±7.0歲）及15名第Ⅲ型SMA患者（平均年齡爲13.7±8.2歲）。以徒手肌力測量法測量每一患者共71個肌群，以取得個別肌群肌力分數，及特定部位：包括位於四肢關節上的肌群、上肢的屈曲肌和伸展肌，及下肢的屈曲肌和伸展肌等之平均肌力分數。使用多變數變異量分析法及成對t檢定以各自分析第Ⅱ型及第Ⅲ型SMA患者之肌力資料。測量結果顯示在兩類型SMA患者中，軀幹及髖關節處的個別肌群肌力分數較低，而手肘屈曲肌、手腕屈曲肌及伸展肌、手指屈曲肌及橫隔膜肌則呈現較高的肌力分數。在第Ⅱ型SMA患者中，位於左側肩胛關節、手肘及手腕關節處之肌群所得之平均肌力分數比在右側該處肌群之平均分數顯著地較低。結果中同時顯示在兩類型SMA患者中，上肢屈曲肌之平均肌力分數顯著地高於上肢伸展肌之平均分數。下肢屈曲肌平均分數顯著地高於下肢伸展肌則只見於第Ⅱ型SMA患者中。從本研究中能了解第Ⅱ型及第Ⅲ型SMA患者之肌肉力量及身體各部位肌力的差異情況。

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